Health-related quality of life among children with mental health problems

Thesis presented to the Faculty of Arts of the University of Zurich for the degree of Doctor of Philosophy

by

Michelle Dey

of Marsens (FR)

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ABSTRACT

BACKGROUND: Health-related quality of life (HRQOL) is a subjective, multidimensional and dynamic construct that encompasses physical, psychological and social function. The present thesis focused primarily on HRQOL among children with mental health problems, because this group has been neglected in HRQOL research to date. **OBJECTIVES**: Prior to empirical investigation, a systematic review of the literature was conducted, reviewing all existing studies on HRQOL among children with various mental disorders, relative to healthy controls, and describing the various limitations of these studies. Subsequently, an empirical, population-based survey was conducted, again studying HRQOL among children with mental health problems, as well as children with physical health problems and healthy controls: (1) to assess the influence of health status and additional health-related predictors on HRQOL; (2) to analyze the effects of item overlap between symptoms of mental health problems and HRQOL measurements; and (3) to evaluate levels of parent-child agreement on a child's HRQOL. **METHODS**: For the systematic literature review, relevant publications were searched using different databases and search terms, as well as by checking reference lists and contacting experts. Articles were included that (1) compared children with mental disorders to healthy controls/norm values or made such comparisons possible; and (2) fulfilled pre-defined inclusion criteria. A population-based cross-sectional survey then was conducted for the empirical component of the present thesis. Children ages 9-14 years and living in Switzerland were identified, recruited and analyzed. Parents and/or children themselves rated the child's HRQOL using the KIDSCREEN-27. A HRQOL assessment ultimately was available for 535 children with mental health problems, 327 children with physical health problems, and 744 healthy controls. **RESULTS**: Literature review revealed that the HRQOL of children with various mental disorders is compromised relative to healthy controls, especially within psychosocial and parent/family-related domains. The most important limitations of existing research includes the lack of population-based studies (samples mostly were drawn from psychiatric clinics), the failure to use self-ratings (many authors only used proxy-ratings), and failure to consider all possible explanations for compromised HRQOL (e.g., item overlap; status of medication use; severity of a child's health problem). In the empirical survey itself, simple regression analyses revealed that both children with mental health problems and those with physical

health problems have compromised HRQOL, relative to healthy controls. However, on multiple regression analysis, the severity of symptoms of mental and physical health problems was the most important predictor of reduced HRQOL. Furthermore, controlling for item overlap between symptoms of mental health problems and HRQOL items did not significantly alter results. Lastly, the level of agreement between proxy- and self-ratings of a child's HRQOL was relatively high. Nevertheless, some parent-child pairs disagreed, with self-ratings often higher than proxy-ratings.

CONCLUSIONS: Children with mental health problems have reduced HRQOL relative to healthy controls, a result that is not (solely) attributable to item overlap between the diagnostic criteria of mental health conditions and HRQOL measurements. This suggests that HRQOL assessments provide information that goes beyond the symptoms of a mental health condition, thereby providing a broader picture of the effects that mental health problems and their treatment have on children. That both mental and physical health conditions are associated with reduced HRQOL emphasizes how problematic the current neglect of mental health problems in HRQOL research is. Furthermore, that the severity of health problems is a very important HRQOL predictor must be considered when the HRQOL of children with mental and physical health problems are compared. Lastly, the finding that parent-child disagreement does exist in HRQOL ratings means that (1) proxy- and self-ratings should both be used, whenever possible; and (2) when only proxy-ratings are obtainable, they should be interpreted as merely the perspective of the parents, which might be influenced by different factors and, thereby, not accurately reflective of the child's perceptions. Even though the current study attempted to overcome many of the limitations of prior research on HRQOL among children with mental health problems, further studies in this research field remain necessary to fill numerous knowledge gaps. Suggestions for subsequent investigations are provided.

ABBREVIATIONS

ADHD: Attention-deficit/hyperactivity disorders

ASD: Autism spectrum disorders

CATI: Computer-assisted telephone interviews

CHIP: Child Health and Illness Profile

CHQ: Child Health Questionnaire

CI: Confidence interval

CSHCN: Children with special health care needs

DSM-IV-TR: Diagnostic and Statistical Manual of Mental Disorders

DUX-25: Dutch-Child-AZL-TNO-Quality-of-Life

ES: Effect sizes

HRQOL: Health-related quality of life

ICC: Intraclass correlation coefficient

ICD-10: International Classification of Disease and Related Health Problems

ISCED: International Standard Classification of Education

KINDL-R: Questionnaire for Measuring Health-Related Quality of Life in Children and Adolescent - Revised Version

PedsQL: Pediatric Quality of Life Inventory

NS-CSHCN-CH: National Survey of Children with Special Health Care Needs in Switzerland

SD: Standard deviation

SDQ: Strength and Difficulties Questionnaire

SNSF: Swiss National Science Foundation

SpLD: Specific learning disabilities

TACQOL: TNO-AZL-Child-Quality-Of-Life

QOL: Quality of life

VIF: Variance inflation factors

WHO: World Health Organization

1 General Introduction

1. GENERAL INTRODUCTION

The aim of the present thesis was to evaluate *health-related quality of life (HRQOL)* among *children* (for simplicity the term children is used for individuals of ages 0 to 18 years). The study on which this thesis is constructed included three groups: 1) children with mental health problems, 2) children with physical health problems, and 3) healthy controls. However, the current work focused specifically on the first group, because children with mental health constraints have been neglected in HRQOL studies, to date.

To distinguish between *mental* and *physical health problems*, this thesis applied the 'International Classification of Disease and Related Health Problems' (ICD-10; [1]) as an analytical framework. Hereby, health constraints that are listed in Chapter V of the ICD-10 ('Mental and behavioral disorders') were labeled as 'mental health problems', whereas health constraints from Chapters I to IV or VI to XIX were treated as 'physical health problems'. Subsequently, the term *'disorder'* is used when the mental health condition of a person was diagnosed in detail (e.g., through a specialist). In contrast, expressions like *'health problems'* or *'health constraints'* are used when no detailed diagnostic information was available about a person (e.g., in the empirical part of the present thesis) or when the influences of such conditions on HRQOL are discussed in a general manner.

The present work consists of five chapters. *Chapter 1* provides a general overview. Section 1.1 defines the HRQOL concept and suggests possible applications of the construct. Subsequently, methodological challenges that must be considered when measuring HRQOL of children and/or individuals with mental health problems are discussed in Section 1.2. Section 1.3 describes HRQOL among adults with mental health problems. Section 1.4 introduces the present study – the 'National Survey of Children with Special Health Care Needs in Switzerland' (NS-CSHCN-CH). *Chapter 2* provides a systematic review of studies that have compared the HRQOL of children with mental disorders with that of healthy controls, followed by a discussion about the limitations of existing studies. The empirical findings of the NS-CSHCN-CH are presented in the third and fourth chapters. *Chapter 3* describes different variables (e.g., health status, severity of symptoms) that contribute to the prediction of HRQOL. Furthermore, the effects of item overlap between the conceptualization of mental health problems and HRQOL measurements are illustrated. *Chapter 4* assesses the level of

agreement between self- and proxy-ratings. Finally, *Chapter 5* offers a general discussion, which includes summarizing and discussing all results (Section 5.1), a description of the strengths (Section 5.2) and limitations (Section 5.3) of the present thesis and original study, suggestions for subsequent research (Section 5.4), and final conclusions (Section 5.5).

1.1 Defining and applying HRQOL

Due to medical advances, biomedical successes, like improved survival rates, have been achieved for individuals with many severe physical health conditions (e.g., cancer [2-7]). However, these successes have not always been mirrored by positive self-evaluations by the patient. Due to this discrepancy, it has become evident that both the *subjective perception* of the patient and a *bio-psychosocial* assessment are necessary to obtain a comprehensive picture of the effect of a health condition and its treatment. This shift from a purely biomedical to a bio-psychosocial perspective has taken place not only in the field of somatic medicine, but also in the field of psychiatry [8] (e.g., due to the recognition that the side effects of antipsychotic drugs are associated with reduced well-being among many patients with schizophrenia [9; 10]).

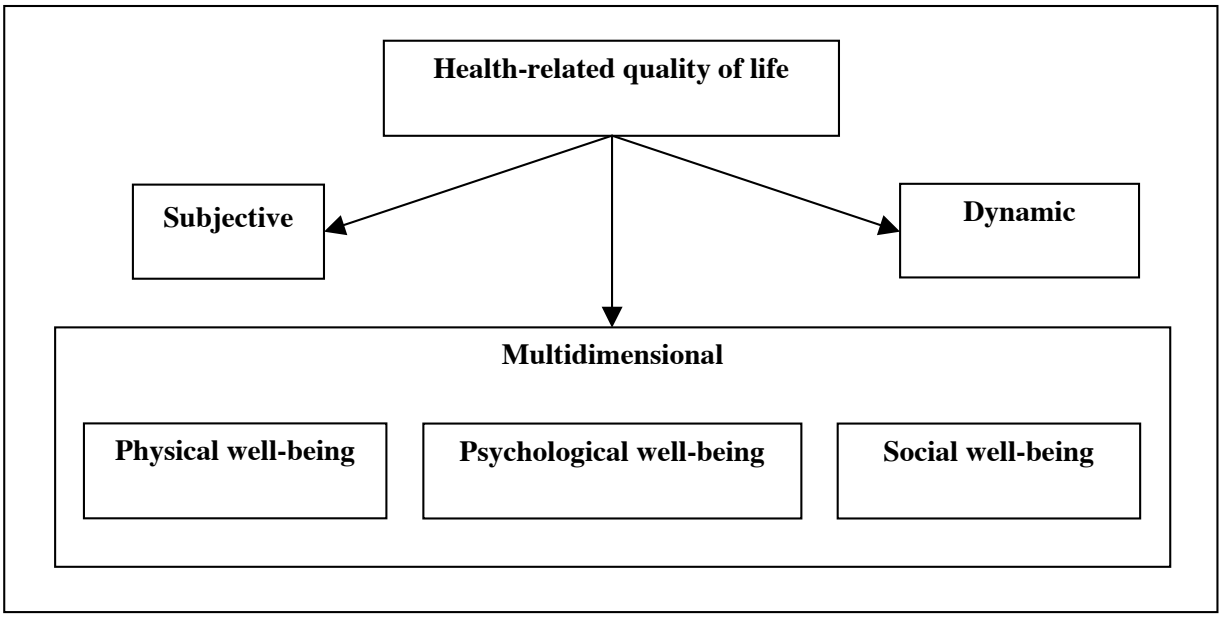
This afore-mentioned shift was already initiated in 1946, when the World Health Organization (WHO) defined health as *'... a state of complete physical, mental, and social well-being and not merely the absence of disease or infirmity'* [11]. This definition emphasizes that health and the effects of health care should also include an estimation of *well-being*. Later on, it was proposed that the concept of *quality of life (QOL)* should be used to assess this subjective and bio-psychosocial well-being [12]. Hence, the WHO definition of health can be interpreted as a precursor to the QOL concept [8].

Even though the importance of QOL has been acknowledged, the lack of a universal valid definition has been criticized (e.g., [13-18]). Furthermore, this concept is frequently not clearly separated from HRQOL [18; 19]. The broader QOL term includes the dimensions of HRQOL (see below) as well as additional dimensions (e.g., political freedom; [20]). Subsequently, the term HRQOL is used because

it is mostly applied when studying individuals with mental or physical health constraints, as in the NS-CSHCN-CH.

Throughout this thesis, the HRQOL definition of Taylor et al. [17] is used, because it is particularly applicable to the situation of the *children with chronic health conditions* targeted in this study. According to these authors, the following three characteristics define the concept (also see Figure 1.1): HRQOL is 1) *subjective* (i.e., it is unique to each individual and depends upon self-evaluation); 2) *multi-dimensional* (i.e., it includes aspects of physical, psychological, and social function and, therefore, corresponds to the above-mentioned bio-psychosocial perspective); and 3) *dynamic* (i.e., HRQOL depends upon the stage of development of the child, the illness trajectory, the achievement of goals, and aspirations, as well as on the constraints imposed through ill-health and treatment). These defining characteristics are also inherent in most other HRQOL definitions.

Figure 1.1: Defining health-related quality of life



Despite the lack of a universal valid definition, the number of HRQOL publications has increased greatly in recent decades [13; 16; 19]. In current times, HRQOL is applied for various purposes (e.g., [13; 18; 19; 21-26]). In the *therapeutic context,* HRQOL assessments are used 1) to obtain a comprehensive picture about the effects of a particular health condition on a person's life; 2) to plan treatment (e.g., relevant themes can be identified and prioritized); 3) to monitor changes during the therapeutic process and, if necessary, adapt the treatment; and 4) to broadly evaluate the success of treatment after its conclusion. However, HRQOL assessments must not be used only at the level of the individual, but also at the *population level* (e.g., to monitor the health status of an entire population, to detect health inequalities, to plan health services, to allocate resources, and to evaluate implemented interventions).

1.2 Assessing HRQOL

Over the past few decades, the number of HRQOL measurements has increased markedly [13; 26]. One must first consider a *generic* or *disease-specific* instrument to select the instrument best suited for a particular research or clinical context (see [19; 22; 25-28]). The advantage of generic instruments is that they can be used for healthy and unhealthy individuals. The disadvantage of such measurements is that they lack sensitivity towards areas that are especially important for individuals with a particular health constraint. Hence, if one aims to assess these condition-specific HRQOL areas or treatment-related changes, disease-specific instruments may be more appropriate.

Additionally, it should be considered that HRQOL instruments should reflect the *multidimensionality* of this concept by including at least physical, psychological, and social functioning (see Section 1.1). Most instruments appear to fulfill this criterion [26; 29]. However, the operationalizations of these super-ordinate HRQOL domains varies considerably across different measurements [8; 22; 26; 29]. Hence, it is difficult to compare HRQOL studies that have used distinct instruments [18].

Lastly, a HRQOL instrument has to be *culturally suitable*, *reliable, valid, sensitive to change* (e.g., in order to assess the effect of interventions) and *brief* [22; 30].

As mentioned previously, this thesis focuses on *HRQOL among children with mental health problems*. Therefore, subsequent sections identify methodological characteristics that must be considered when children and/or individuals with mental health problems are studied. In the present thesis, generic measurements are relevant because various health groups are compared (see Section 1.4.1.1.3). An overview about selected generic HRQOL measurements that can be used to assess HRQOL among children is provided in Table 1.1. Most of these instruments have been used frequently. More details about this instruments are offered in Chapter 2 to 4.

Table 1.1: Selected generic measurements to assess health-related quality of life among children

Measurement (Abbreviation)	**Author**	**Items**	**Age range**	***Total HRQOL score*/scales**/subscales [a]
Child Health and Illness Profile (CHIP)	Riley et al. [31]	*Self-report*: child version: 45; adolescent version: 183 *Proxy-report*: 76	*Self-report*: child version: 6-11; adolescent version: 12-17 *Proxy-report*: 6-11	Achievement; Risk avoidance; Satisfaction; Resilience; Comfort; Discomfort
Child Health Questionnaire (CHQ)	Landgraf et al. [32]	*Self-report*: 87 *Proxy-report*: different versions with 28, 50 or 98 items	*Self-report*: 10-18 *Proxy-report*: 5-18	**Psychosocial Health; Physical Health** Role/social limitations-emotional; Role/social limitations-behavioral; Behavior[b]; Mental health; Self-esteem; Parent impact-emotional[c]; Parent impact-time[c]; Family activities; Family cohesion[b]; Physical functioning ; Role/social limitations-physical; Bodily pain/discomfort; General health perceptions; Change in health
Dutch-Child-AZL-TNO-Quality-of-Life (DUX-25)	Kolsteren et al. [33]	*Self- and proxy-report*: 25	*Self- and proxy-report*: 5-16	***Total HRQOL score*** Home; Physical; Emotional; Social

Measurement (Abbreviation)	**Author**	**Items**	**Age range**	***Total HRQOL score*/scales**/subscales [a]
KIDSCREEN	Ravens-Sieberer et al. [24]	*Self- and proxy-report*: different versions with 10, 27 or 52 items	*Self- and proxy-report*: 8-18	***Total HRQOL score*** Physical well-being; Psychological well-being; Moods & emotion; Self-perception; Autonomy; Parent relations & home life; Social support & peers; School environment; Social acceptance (bullying); Financial resources
Questionnaire for Measuring Health-Related Quality of Life in Children and Adolescent - Revised Version (KINDL-R)	Ravens-Sieberer & Bullinger [34]	*Self-report*: 4-to-7- year-olds: 12; 8-to-6-year-olds: 24 *Proxy-report*: 24	*Self-report*: 3 age versions -> 4-7; 8-12; 13-16 *Proxy-report*: 2 age versions -> 4-7; 8-16	***Total HRQOL score*** Friends; Family; Self-esteem; School; Emotional well-being; Physical well-being
Pediatric Quality of Life Inventory (PedsQL)	Varni et al. [35; 36]	*Self- and proxy-report*: 23	*Self-report*: 3 age versions -> 5-7; 8-12; 13-18 *Proxy-report*: 4 age versions -> 2-4; 5-7; 8-12; 13-18	***Total HRQOL score*** **Psychosocial Health Summary Score**; **Physical Health Summary Score**[d] School Functioning; Emotional Functioning; Social Functioning; Physical Functioning[d]
TNO-AZL-Child-Quality-Of-Life (TACQOL)	Verrips et al. [37-39]	*Self- and proxy-report*: 56	*Self-report*: 8-15	Cognitive functioning; Social functioning; Motor functioning; Autonomic functioning; Bodily functioning; Negative moods; Positive moods

Note: *HRQOL*: health-related quality of life

[a] Always the (sub)scales of the longest version are mentioned. Sometimes, some of the mentioned subscales are merged in the shorter versions.

[b] Only consists of one item

[c] only computable in the parent's version

[d] The 'physical health summary score' contains the same items as the subscale 'physical functioning'.

1.2.1 Measuring HRQOL in children

It is important to evaluate the HRQOL of children separately from adults, because certain issues are specific to this age group [15; 19; 28; 40; 41]. For instance, children undergo impressive physical and psychosocial development, face other developmental tasks, life events and stressors, participate in other contexts (e.g., school), and depend more on other people. Furthermore, children and adults differ regarding the prevalence and manifestations of particular health conditions. However, HRQOL research focusing on children represents a relatively new field [20; 22]. Bullinger and Ravens-Sieberer [42] estimated that only about 13% of all HRQOL publications target children. Many of these publications are limited, because they merely describe a particular HRQOL instrument without offering any detailed investigation of HRQOL in children with specific health conditions.

1.2.1.1 Age of the child

Since children are often perceived as unreliable respondents, some authors have questioned whether children should ever self-rate their HRQOL (see [22]). However, more recent publications have emphasized that children should also self-rate their HRQOL, whenever possible [28]. In order to do so, the child has to 1) understand the questions (if a written and self-administered version is used, the child has to possess the necessary reading skills) and must be able to respond in a given answer format; 2) be able to refer his/her answer to the time frame that is prompted in the HRQOL measurement (e.g., the past week); and 3) be able to maintain attention over the time that is necessary to complete the questionnaire [18; 28; 43]. However, children develop these skills at different ages [19]. For instance, the understanding of written HRQOL questions may be delayed in children with learning disabilities [27]. Due to these developmental variations, it is difficult to formulate a general rule about the specific age at which a child is able to appropriately self-rate his/her HRQOL. Nevertheless, even very young children (around 4 years of age) can often provide some information on concrete aspects of their health [28]. In contrast, their ability to rate more subjective and complex HRQOL domains develops later [28]. Summarizing various studies, it can be concluded that children ≥ 8 years are generally able to understand HRQOL questions and answer them in a reliable and valid

manner [43; 44]. However, it is important to determine the lower age limit for self-ratings separately for each HRQOL measurement [28].

1.2.1.2 Proxy-ratings

Despite the importance of self-ratings, *proxy-ratings* provide important additional information and should be considered in addition to self-ratings [19]. Furthermore, in some cases (e.g., when the child is too young or cannot self-rate his/her HRQOL due to suffering from a particular health condition), proxy-ratings are the only way to assess the HRQOL of the child [18; 19]. A detailed discussion of the importance of using self- and proxy-ratings, as well as a description of agreement between self- and proxy-ratings, is provided in Chapter 4. At this point, it is important to highlight that the child and his/her parents should be given parallel versions of a HRQOL questionnaire to rate the child's HRQOL [22].

1.2.1.3 HRQOL contents

The age of the child is associated not only with the skills that are necessary for them to self-rate their HRQOL, but also with the *contents* that are perceived by the child as being relevant [18; 28]. Hence, HRQOL items should be formulated broadly to make their content applicable to children across a wide range of ages. Such an approach is advantageous in that the same instrument can be used to assess HRQOL among children of different ages. However, the disadvantage of such an approach is that it is likely to miss important information about age or developmentally-specific HRQOL [18]. Regarding the content of HRQOL measurements, it is further important to consider the perspective of children not only during the HRQOL assessment, but also during the creation of HRQOL instruments. That is, the children's opinion about what constitutes HRQOL and what domains are important for them must be integrated into the development of specific items, (sub)scales, and entire HRQOL measurements. To date, this has rarely been done [18; 42].

1.2.2 Measuring HRQOL in individuals with mental health problems

HRQOL research has generally paid less attention to individuals with *mental health problems* than those with *physical health constraints* [19; 21; 26; 28; 30; 43]. This negligence is regrettable, because mental disorders are frequent phenomena in children, as well as in adults, and are often associated with long-lasting negative effects (e.g., [30; 45-48]). This lack of attention to the relationship between *mental disorders* and *HRQOL* can be explained partially by the methodological challenges (see subsequent sections) that arise when studying this particular group.

1.2.2.1 Item overlap

Item overlap is defined as contentual similarities between HRQOL items and items that are utilized to assess the presence of a particular health constraint [8; 27]. In psychosocial HRQOL domains, this problematic item overlap is greater for mental (especially for anxiety disorders and depression) than physical health problems [8; 19; 27; 30]. For instance, some of the HRQOL questions of the *'World Health Organization Quality of Life'* questionnaire [49] (e.g., 'How much do you enjoy life?'; 'Have you been able to concentrate?'; 'How satisfied are you with yourself?') are strongly related to the symptoms of depressive episodes (e.g., decreased mood, reduced concentration, as well as reduced self-esteem and self-confidence) [1]. Hence, it can be argued that measuring HRQOL is tautological when psychopathological symptoms and HRQOL items are too redundant [8]. However, despite this item overlap, various authors emphasize that a HRQOL assessment is a useful addition to the assessment of psychopathological symptoms, at least when conceptualized broadly and multi-dimensionally (e.g., [15; 19; 30]). Nevertheless, researchers should control for item overlap in their statistical analyses or at least consider this effect when interpreting the results of studies conducted with individuals with mental health problems [8; 27; 30].

1.2.2.2 Psychopathological fallacies

The second challenge in measuring HRQOL among people with mental disorders involves three *psychopathological fallacies* (see [8; 19; 27; 30]). 1) The *affective fallacy* arises because people's

judgments of their well-being are based on their current affective state. Hence, depressed patients rate their HRQOL as overly negative, whereas patients with mania exhibit the opposite bias. 2) The *reality distortion fallacy* occurs when experts take the HRQOL reports of people with symptoms of delusions and hallucinations for granted, even though such psychopathological symptoms distort these ratings. 3) The *cognitive fallacy* arises when the self-rated HRQOL of intellectually-impaired individuals (e.g., people with mental retardation) is interpreted as valid. Due to the existence of these three fallacies, it is important to evaluate the HRQOL of patients with mental health constraints not solely based on self-ratings, but also on the ratings of relevant others (e.g., family members) [50].

1.3 HRQOL of adults with mental health problems

This section provides a review of HRQOL among adults with mental disorders (for an extensive overview, see [51]). A detailed description of HRQOL among children with mental health problems is provided in Chapters 2 through 5.

Different reviews emphasize that adults who suffer from one of the two most prevalent mental disorders – i.e., *anxiety* or *mood disorders* [52; 53] – have reduced HRQOL relative to healthy controls (anxiety disorders [54; 55]; bipolar disorders [56]; major depressive disorders [57]). Furthermore, their HRQOL is similar to or more compromised than HRQOL among individuals with physical health conditions. However, it is possible that item overlap (see Section 1.2.2.1) and/or the affective fallacy (see Section 1.2.2.2) can explain these results, at least partly [8; 58]. Nevertheless, additional explanations also must be considered for studies that define HRQOL broadly. For instance, Schneider and Pantol [55] proposed that the compromised HRQOL in adults with anxiety disorders is due not only to particular characteristics of such health conditions (e.g., distress with experiencing anxiety; avoidance behavior), but also to the stigma associated with such mental disorders.

Compromised HRQOL compared to the general population and compared to people with various physical health constraints has also been reported among patients with *schizophrenia* [10]. Such an ample effect on HRQOL is not surprising, since schizophrenia has been declared one of the most

burdensome and costly health conditions worldwide [47]. HRQOL seems to be particularly compromised in schizophrenic patients, due to the symptoms of this mental disorder (especially negative, cognitive and depressive ones) as well as adverse events associated with treatment [10]. However, the *reality distortion fallacy* (see Section 1.2.2.1) has to be considered when patients suffering from schizophrenic symptoms self-rate their HRQOL.

Childhood-onset mental health problems can also negatively affect the HRQOL of adults. For instance, previous studies have demonstrated that adults with a *high functioning autism-spectrum disorder* exhibit lower HRQOL than healthy controls [59; 60]. Thereby, not only the social HRQOL domain, which is closely related to the core symptoms of such disorders, was affected, but also additional HRQOL domains (e.g., physical well-being). Hence, the HRQOL constraints of such patients cannot be attributed solely to the effects of item overlap. Other mental disorders with an early onset (e.g., *attention-deficit/hyperactivity disorder* (ADHD) [61], *learning disabilities* [62], *specific language impairments* [63]) also seem to compromise HRQOL in adults.

In summary, adults with various mental disorders report lower HRQOL than healthy individuals and a comparable or lower HRQOL than many individuals with physical health constraints. Given these observations, the following issues must be considered: 1) The methodological challenges associated with studying HRQOL among people with mental disorders (see Section 1.2.2) must be addressed to improve the interpretation of these results. 2) However, as mentioned above, compromised HRQOL in individuals with mental disorders cannot be attributed solely to methodological characteristics (e.g., item overlap). Hence, additional explanations for the marked reductions in HRQOL in individuals with mental health constraints have to be considered as well. 3) The described results apply at a group level. Hence, it is possible that the HRQOL of some individuals with mental disorders is not compromised. Accordingly, Katschnig [8] described how many people with long-lasting mental disorders are satisfied with life conditions that would be regarded as inadequate by external standards. This effect arises because such patients adapt to their situation by lowering their standards regarding their HRQOL rating [8]. In other words, the presence of a particular health condition can lead to changes in 1) the person's internal standards; 2) his/her values (i.e. the importance of particular domains, which constitute the target construct); or 3) his/her conceptualization of the specific

construct. These changes subsequently modify the evaluation of the particular construct, for instance to an improved HRQOL rating [64]. This adaptive process is called *'response shift'* [64] and was so far predominantly discussed for people with life-threatening or chronic physical health conditions (e.g., cancer; [65]). Studies about the effects of a response shift on HRQOL ratings among individuals with mental disorders are sparse. Hence, even though Evans et al. [66] concluded that response shift does not have a large effect on self-rated HRQOL among people with mental disorders, this issue needs to be investigated further.

1.4 Present study

The present thesis is based on the Swiss National Science Foundation (SNSF) project entitled '*Children with special health care needs (CSHCN) in Switzerland: Prevalence, health care utilization and social determinants'* (project number: 325130_125486). According to the definition of the Maternal Child and Health Bureau, CSHCN were defined as *'...those who have or are at increased risk for a chronic physical, developmental, behavioral or emotional condition and who also require health and related services of a type or amount beyond that required by children generally'* [67].

The SNSF project was initiated by Meichun Mohler-Kuo (Institute of Social and Preventive Medicine, University of Zurich). The four main aims of the project were:

1. To estimate the prevalence and to describe the characteristics of CSHCN in Switzerland using a national representative sample;
2. To evaluate HRQOL and access to health care services among CSHCN in Switzerland;
3. To identify multi-level indicators associated with health care service use among CSHCN;
4. To compare the prevalence and characteristics of CSHCN in Switzerland with data drawn from a similar national survey conducted in the United States.

The author of the present thesis was employed as a research fellow conducting the study under the supervision of Meichun Mohler-Kuo. The Ph.D. student made crucial contributions to study design

(e.g., development of the questionnaires), was responsible for project implementation (e.g., corresponding with the data protection officers, municipalities/cantons and parents; organizing the translations, printing and mailing the study material; cooperating with the institute that conducted the telephone interviews; and drafting annual reports for the SNSF), and processed data (data entry; data cleaning; data analyses). Furthermore, she was responsible for writing three journal articles (see Chapters 2 to 4).

1.4.1 Methods

1.4.1.1 Study design

The NS-CSHCN-CH was conducted between 2010 and 2011. The ethics committee of the Canton of Zurich and all data protection officers approved the study protocol. The subsequent sections illustrate the sampling method and the two phases of the survey. An overview of the measurements and questions used is provided in Table 1.2. The measurements and questions that were especially important for the present thesis are described in greater detail in Chapters 3 and 4.

1.4.1.1.1 Sampling

We intended to obtain a nationally representative sample of children between the ages of 9 and 14 years and living in Switzerland (see Figure 1.2). We chose children younger than 15 years of age, as most large-scale health surveys in Switzerland have targeted respondents 15 years old or older (for instance, the Swiss Health Survey). Furthermore, selecting this age group, as opposed to much younger children, allowed us to obtain a HRQOL assessment both from parents[1] and children (see Section 1.2.1.1).

It was aimed to recruit approximately 1,200-1,500 CSHCN because the targeted number of participants would give us enough power to conduct comparisons among different subgroups (e.g.,

[1] For simplicity, the term *parents* is generally used throughout the thesis, even though sometimes other relevant primary care takers participated in the survey.

subgroups based on socio-demographic characteristics, language regions, health status, etc.). To reach this goal, the targeted sample consisted of 18,000 children. Based on experiences of the pilot project, it was assumed that 90% of all cantons/municipalities would provide the required personal data about 16,200 children (see below). Furthermore it was expected that the response rate would approximately be 60% (corresponds to 9,720 children). Lastly, according to the literature and studies from the U.S. (e.g., [68]), it was estimated that about 15% of all children would meet the criteria of special health care needs (i.e., 1,458 children).

Sampling differed as a function of whether a particular canton possessed a central registry of residents. *Cantons with a central registry (Basel-Landschaft, Basel-Stadt, Berne, Neuchâtel, Geneva, Ticino)* were asked to randomly select a predetermined number of children from their registry who met our age criteria (year of birth: 1996-2000) and send us information about these children (first and last name, birth date, sex, address, nationality) and their parents (first and last names). Two-stage sampling was applied for the remaining *cantons without a central registry* (for details about sampling, see [69]). In the first sampling stage, 252 municipalities were selected randomly. In the second stage, the selected municipalities, analogous to cantons with a central registry, were asked to draw a random sample of a predetermined number of children from their registry and to send us the required personal data. The number of children who had to be sampled varied between 1 and 2,171, depending on the size of the canton or municipality.

As illustrated in Figure 1.2, five municipalities and 130 children had to be excluded from the targeted sample (e.g., because the selected municipality did not exist anymore). After this exclusion, the adjusted targeted sample consisted of 17,870 children from six cantons with a central registry and from 247 municipalities within the remaining cantons. All six cantons as well as 219 municipalities (response rate = 88.9%) sent us the requested information, yielding a total of 16,814 children. During data collection, 318 additional children had to be excluded because the personal data that we received from the cantons/municipalities turned out to be invalid (e.g., because the age criterion was not met or because the address provided was no longer correct). Hence, we obtained valid data for 16,496 children.

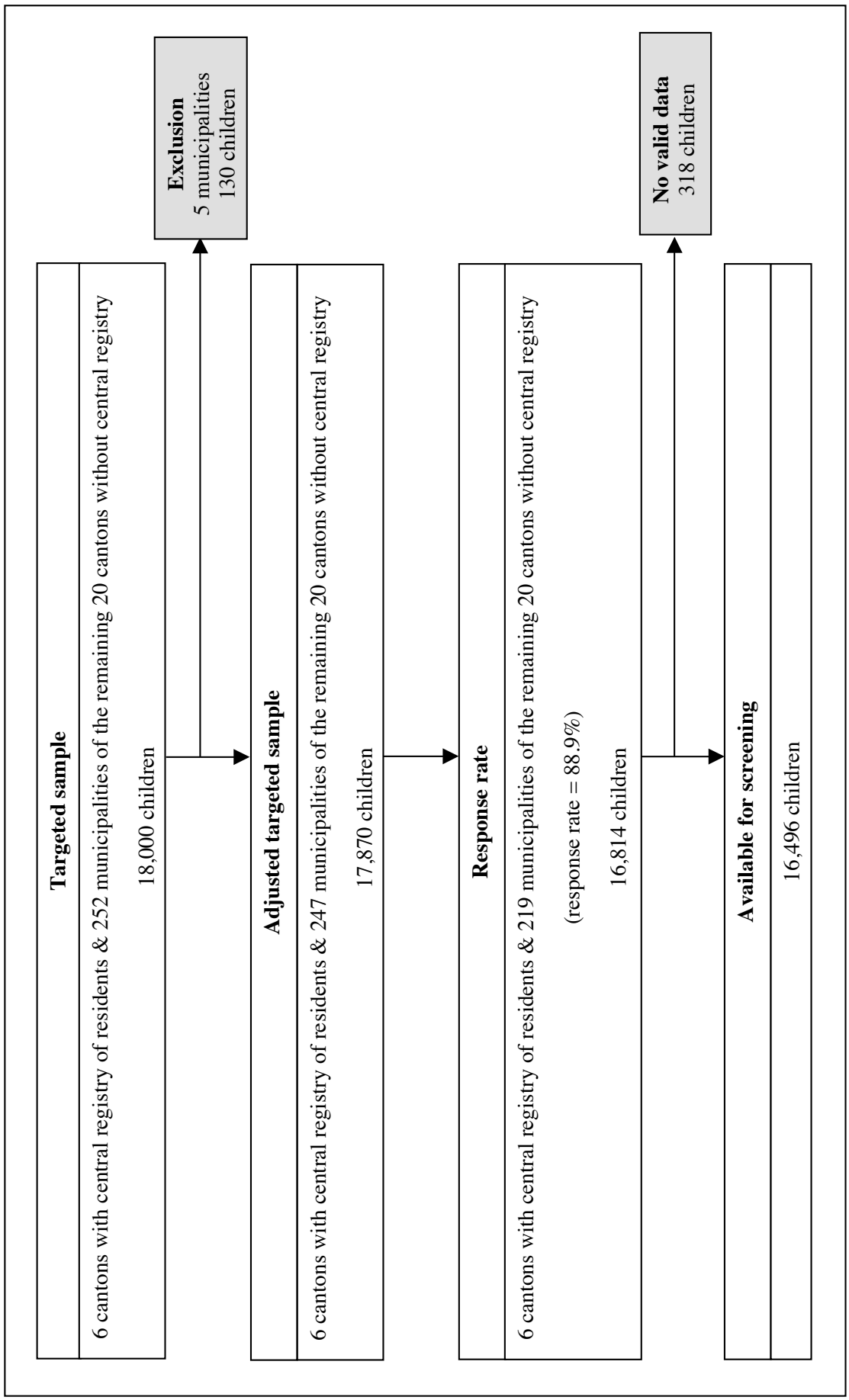


Figure 1.2: Sampling

1.4.1.1.2 Phase I

The main goal of phase I was to screen children for special health care needs using the *CSHCN Screener* (see Figure 1.3 and 1.4; Table 1.2). Screening was conducted via computer-assisted telephone interviews (CATI) with the parents. However, when telephone numbers were not available (2,859 parents), or when parents could not be reached by telephone due to an invalid telephone number or simply due to not responding (total 1,340 parents), the screening questionnaire was sent to parents by mail. On this written questionnaire, the parents were additionally asked to include their telephone number if they agreed to be contacted again for the CATI. Before the telephone interview or accompanying written questionnaire, the parents received a letter introducing different aspects of the study (e.g., aims, voluntary nature of participation, anonymity of data analyses). One reminder including the questionnaire was sent to parents who did not return the written questionnaire within approximately one month.

Figure 1.3: Children with Special Health Care Needs Screener

1. Does your child currently need or use **medicine prescribed by a doctor** (other than vitamins)?
   - ☐ Yes -> Go to Question 1a
   - ☐ No -> Go to Question 2

   1a. Is this because of ANY medical, behavioral or other health condition?
   - ☐ Yes -> Go to Question 1b
   - ☐ No -> Go to Question 2

   1b. Is this a condition that has lasted or is expected to last for *at least* 12 months?
   - ☐ Yes
   - ☐ No

2. Does your child need or use more **medical care, mental health or educational services** than is usual for most children of the same age?
   - ☐ Yes -> Go to Question 2a
   - ☐ No -> Go to Question 3

   2a. Is this because of ANY medical, behavioral or other health condition?
   - ☐ Yes -> Go to Question 2b
   - ☐ No -> Go to Question 3

   2b. Is this a condition that has lasted or is expected to last for *at least* 12 months?
   - ☐ Yes
   - ☐ No

3. Is your child **limited or prevented** in any way in his or her ability to do the things most children of the same age can do?
   - ☐ Yes -> Go to Question 3a
   - ☐ No -> Go to Question 4

   3a. Is this because of ANY medical, behavioral or other health condition?
   - ☐ Yes -> Go to Question 3b
   - ☐ No -> Go to Question 4

   3b. Is this a condition that has lasted or is expected to last for *at least* 12 months?
   - ☐ Yes
   - ☐ No

4. Does your child need or get **special therapy**, such as physical, occupational or speech therapy?
   - ☐ Yes -> Go to Question 4a
   - ☐ No -> Go to Question 5

   4a. Is this because of ANY medical, behavioral or other health condition?
   - ☐ Yes -> Go to Question 4b
   - ☐ No -> Go to Question 5

   4b. Is this a condition that has lasted or is expected to last for *at least* 12 months?
   - ☐ Yes
   - ☐ No

5. Does your child have any kind of emotional, developmental or behavioral problem for which he or she needs or gets **treatment or counseling**?
   - ☐ Yes -> Go to Question 5a
   - ☐ No

   5a. Has this problem lasted or is it expected to last for *at least* 12 months?
   - ☐ Yes
   - ☐ No

We received screening information on about 10,830 children (response rate = 65.7%). Altogether, 9,371 parents completed the CATI, 1,044 filled out the written questionnaire, and 415 completed the written questionnaire before responding to the CATI. The CATI respondents were asked about their relationship to the child of interest. Most often, mothers responded to the interview (78.7%), followed by fathers (20.1%). All other respondent categories (e.g., step parents, adoptive parents, grandparents) were represented with a frequency of < 1%.

No significant sex (X^2_1=2.57, p=.11) or age ($t_{16,494}$=-0.97, p=.33) differences were identified between children of participating and non-participating parents. However, parents of non-Swiss children participated less frequently than parents of Swiss children (X^2_1=454,797, p<.001). This non-participation bias may have been partly due to language/comprehension problems that hindered the participation of some parents of non-Swiss children (see Appendix A.1; [70]). The reasons for refusal to participate are described in Appendix A.1 [70].

Based on the screening, 1,492 children were classified as CSHCN and 9,294 as children without special health care needs (controls); 44 children could not be classified, due to missing data. The latter were excluded from further analyses.

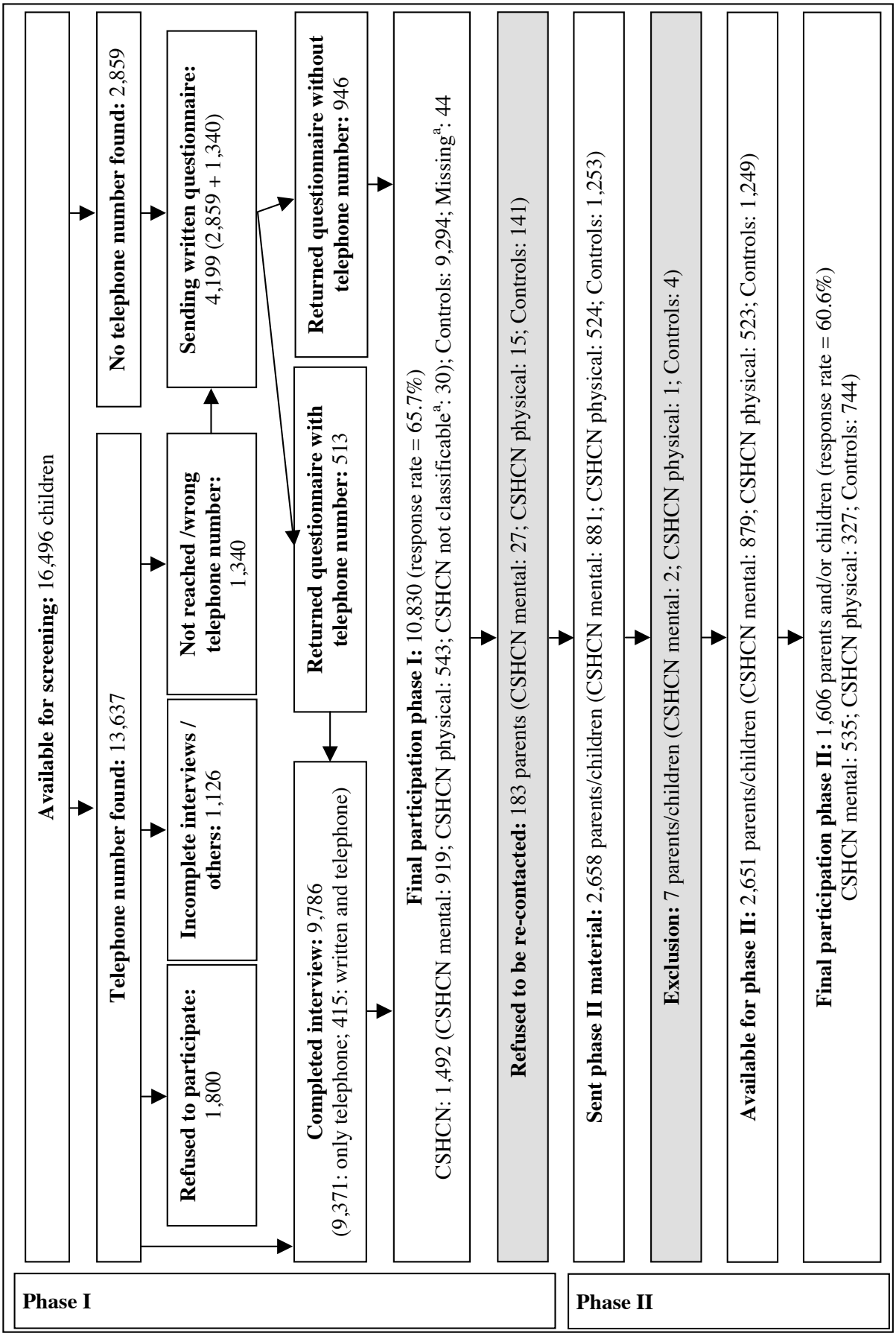


Figure 1.4: Phase I and phase II

Note: CSHCN mental: children with special health care needs with mental health problems; *CSHCN physical*: children with special health care needs with physical health problems

[a] Excluded from the analyses

Table 1.2: Content of the telephone interview/questionnaires of phase I and phase II

Topic	**Items**[a]	***Measurement (reference)* or description of self-developed questionnaire components**[b]	**Respondents**[c] **(mail; tel.)**
Phase I			
Study participation	1	Self-developed: Agreement to participate (= informed consent)	All parents that were reached (tel.)
Refusal	1	Self-developed: Reasons for the refusal to participate	All refusing parents (tel.)
Control questions	2	Self-developed: Child's sex; relationship of the respondent to the child	All (including refusing) parents (tel.)
Special health care needs	14	*CSHCN Screener [71]:* This instrument was developed with the aim to operationalize the definition of CSHCN from the Maternal Child Health Bureau (*'CSHCN are those who have or are at increased risk for a chronic physical, developmental, behavioral or emotional condition and who also require health and related services of a type or amount beyond that required by children generally'* [67]). However, only the children with existing special health care needs are assessed, whereas the *at-risk* population are not captured with the CSHCN Screener.	All parents (mail; tel.)
Neighborhood	5	Selected items about *social cohesion* (feeling of belonging together in the neighborhood) and *social networks* (level of every day interaction among neighbors) of the '*Neighborhood Characteristics Scale*' [72; 73]	All parents (tel.)
Main health problem	1	Self-developed (adapted from the CHQ); [32]): Main health problem of CSHCN	All parents of CSHCN (mail; tel.)
Severity	1	Self-developed (adapted from [74; 75]): Severity of the main health problem	All parents of CSHCN (mail; tel.)
Stability	1	Self-developed (adapted from [74; 75]): Stability of the severity of the main health	All parents of CSHCN (mail; tel.)

Topic	**Items**[a]	***Measurement (reference)* or description of self-developed questionnaire components**[b]	**Respondents**[c] **(mail; tel.)**
Additional health problems	1	Self-developed: Additional health problems that exist beside the main health problem	All parents of CSHCN (mail; tel.)
Physical limitations	1	Self-developed (adapted from the CHQ; [32]): Limitations in schoolwork or activities with friends due to physical health problems	All parents of CSHCN (mail; tel.)
Health literacy	4	Self-developed (adapted from [76; 77]): Knowledge about the health condition of the child and about possible treatments; knowledge about/skills regarding the required care-giving; understanding of information provided in inserts	All parents of CSHCN (tel.)
Health care	44	Self-developed: (Unmet) needs of the child regarding particular health care services; burdens that are associated with the health condition of the child (e.g., financial problems)	All parents of CSHCN (tel.)
Satisfaction with health care	2	Self-developed: Satisfaction with health care services and ease of using them	All parents of CSHCN (tel.); parents of 'extended controls'[d] (tel.)
Barriers to care	36	*Barriers to Care Questionnaire (BCQ; [78]):* Assesses parent-reported experiences/circumstances that may obstruct access to or use of health care, that may reduce the value of clinical encounters, or that interfere with adhering to medical instructions (5 subscales: 'skills', 'pragmatics'', 'knowledge and beliefs', 'expectations', 'marginalization')	All parents of CSHCN (tel.)
Mental health, parents	5	*Mental Health Inventory (MHI-5; [79; 80]):* Assesses mental health status	All parents of CSHCN (tel.); parents of 'extended controls'[d] (tel.)

Topic	**Items**[a]	***Measurement (reference)* or description of self-developed questionnaire components**[b]	**Respondents**[c] **(mail; tel.)**
Physical health, parents	7	*'Physical health'-subscale* from the *World Health Organization Quality of Life-BREF [49]:* Assesses physical health indicators like activities of daily living, energy and fatigue, pain and discomfort etc.	All parents of CSHCN (tel.); parents of 'extended controls'[d] (tel.)
Demographic characteristics	7	Self-developed: Nationality of the child, mother, and father; the number of years parents reside in Switzerland; the highest degree of qualification that the mother/father achieved	All parents of CSHCN (mail; tel.); parents of 'extended controls'[d] (tel.); all parents of controls (mail)
Living situation	8	Self-developed: Living situation of the child (e.g., where/with whom the child lives)	All parents of CSHCN (tel.); parents of 'extended controls'[d] (tel.)
Family	6	Self-developed: (adapted from the CHQ; [32]): Concerns of the parents about the health condition of the child; limitations in the amount of time that the parents have for themselves due to the child's health condition; interference with family activities due to the child's condition; family's ability to get along	All parents of CSHCN (mail; tel.); parents of 'extended controls'[d] (tel.)
Insurance	10	Self-developed: Questions about financial expenditures / benefits through insurance; problems with different kinds of insurance	All parents of CSHCN (tel.)
Further contact	2	Self-developed: Questions about whether the parents agree to be re-contacted for phase II or for the planned long-term survey	All parents (mail, tel.)

Topic	Items[a]	*Measurement (reference)* or description of self-developed questionnaire components[b]	Respondents[c] (mail; tel.)
Phase II			
HRQOL child	27	*KIDSCREEN-27 [24]:* Assesses HRQOL and contains 5 subscales ('physical well-being', 'psychological well-being', 'autonomy & parent relation', 'social support & peers' and 'school environment'). Furthermore, a total HRQOL score can be calculated (based on 10 items)	Parents/children who participated in phase II
Mental health, children	33	*Extended version of the Strength and Difficulties Questionnaires (SDQ; [81; 82]):* Assesses mental health symptoms, positive attitudes, and the consequences of perceived health problems of the child (5 subscales: 'emotional symptoms scale', 'conduct problem scale', 'hyperactivity scale', 'peer problems scale', 'prosocial scale'; a 'total difficulties score'; an 'impact score')	Parents/children who participated in phase II
Relationship satisfaction	7	*Relationship Assessment Scale (RAS; [83; 84]):* Assesses relationship satisfaction (e.g., between the parents and the child)	Parents who participated in phase II
Participation of the child	2	Self-developed: The parents were asked whether or not their child filled out the questionnaire and about the reasons for their child choosing not to participate	Parents who participated in phase II

Note: CHQ: Child Health Questionnaire; *CSHCN:* children with special health care needs; *HRQOL*: health-related quality of life; *tel.*: telephone

[a] Number of items, which also includes sub-questions. In each case, the maximum number of items is indicated. Normally, fewer questions were asked due to the employment of filter questions. Sometimes, not an entire measurement was used due to time constraints.

[b] A detailed description of all measurements or self-created variables relevant to the present thesis is provided in subsequent chapters.

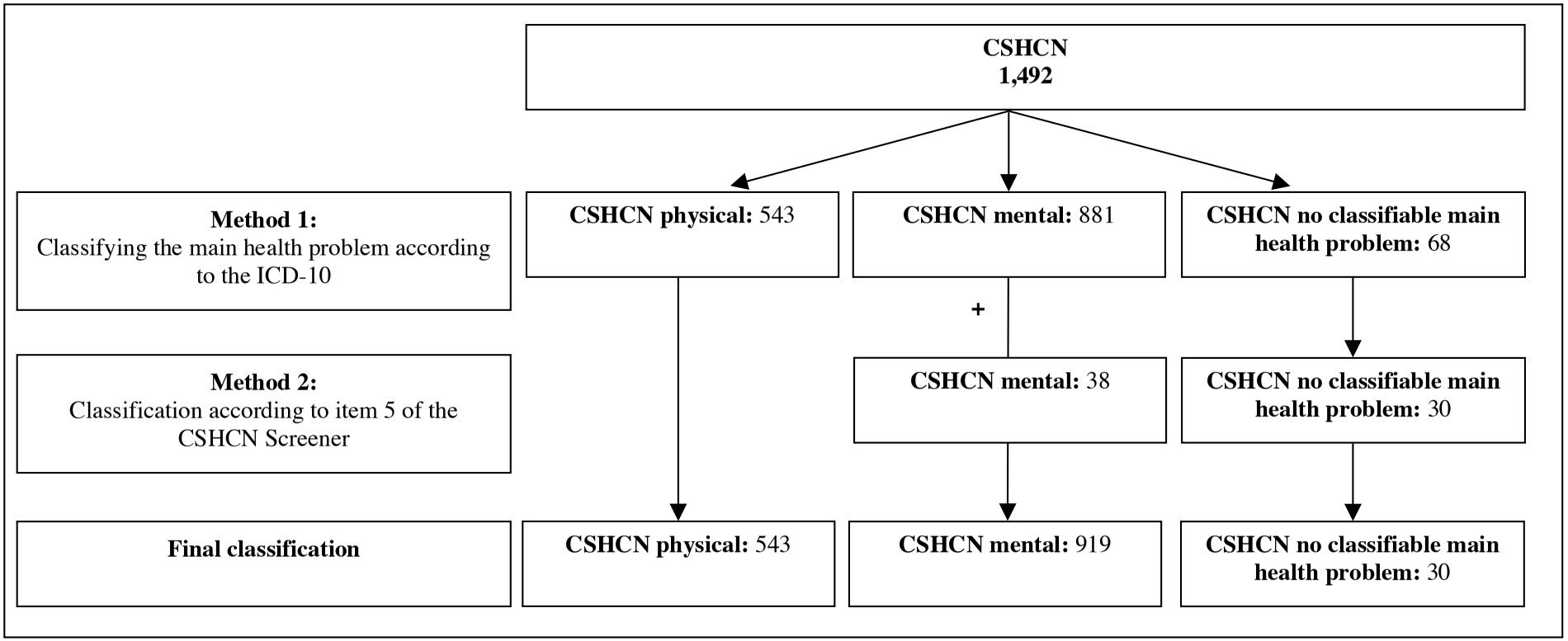
[c] When not otherwise indicated, 'respondents' only refers to participating parents.

[d] We intended to conduct an extended telephone interview with about 1,200 randomly-selected controls. The parents of the non-selected controls had to answer only the *control questions*, the questions about *special health care needs* and *neighborhood*, and the questions about *further contact*.

As described in Table 1.2, additional measurements/self-developed items were used during phase I. The particular questions varied as a function of the health status of the child (whether or not he/she had special health care needs) and research mode (telephone versus mail). For parents of healthy controls who were reached by telephone, the questions depended on whether they were randomly selected for the extended interview.

Based on the additional phase I information and due to the aims of the present thesis, CSHCN were further divided into 1) *CSHCN with mental health problems* (*N*=919), 2) *CSHCN with physical health problems* (*N*=543), and 3) *CSHCN with no classifiable main health problem* (*N*=30). This group assignment was accomplished using two methods (see Figure 1.5). First, the parent-reported *main health problem* (open answer format) of CSHCN was coded according to the ICD-10 [1]. Children with the main health problem belonging to Chapter V ('Mental and behavioral disorders') were assigned to the group *CSHCN with mental health problems*, whereas children with a main health problem belonging to Chapters I to IV or VI to XIX were assigned to the group *CSHCN with physical health problems*. Altogether, 68 CSHCN could not be assigned to either one of these groups (e.g., because the parents did not report a specific health problem) using this method. A second method was therefore applied to classify these individuals. That is, if item 5 of the CSHCN Screener was positive (indicating the need for treatment or counseling for emotional, developmental, or behavioral problems), the children were assigned to *CSHCN with a mental health problem* (see [85; 86]). With this second method, an additional 38 children became classifiable. The remaining 30 CSHCN with no classifiable main health problem were excluded from further analysis. Of the CSHCN with classifiable main health problems and controls, 183 parents refused to be re-contacted for phase II (see Figure 1.4).

Figure 1.5: Classification of children with special health care needs



Note: *CSHCN mental*: children with special health care needs with mental health problem; *CSHCN physical*: children with special health care needs with physical health problems

1.4.1.1.3 Phase II

The main goal of phase II was to obtain information about self- and proxy-rated HRQOL among all CSHCN and a group of randomly-selected controls. Regarding CSHCN, only classifiable participants (see preceding section) were of interest for the present thesis and are considered subsequently.

HRQOL was assessed using the *KIDSCREEN-27* [24]. This instrument was chosen because it has many advantages compared to other available HRQOL measurements. 1) The KIDSCREEN-27 is a generic instrument and therefore enables a comparison of healthy and unhealthy children and children with different health constraints. 2) This instrument measures multiple dimensions of HRQOL because it contains five HRQOL subscales (see Table 1.2). 3) Switzerland was included in the development phase; hence, cultural appropriateness should be guaranteed. 4) The KIDSCREEN-27 is relatively brief. 5) There are two parallel versions of this instrument – one that can be completed by children and one by parents. 6) Focus groups with children were conducted during the development phase of the KIDSCREEN-27; thus, the meaning of HRQOL from the perspective of the child was incorporated. Other instruments that were used in phase II are described in Table 1.2.

Figure 1.4 depicts that 2,658 HRQOL questionnaires were sent out immediately after screening. Some of the CSHCN were not re-contacted for phase II, even though the parents did not refuse further contact. This was because these parents returned the questionnaire from phase I after the time window for the mailing of the phase II questionnaire had already closed. Furthermore, seven parent-child pairs had to be excluded because they were no longer contactable. We received completed questionnaires from 60.6% of the parents and/or children (1,606) of the remaining 2,651 parent-child pairs. Hence, we had HRQOL data for about 535 CSHCN with mental health problems, 327 CSHCN with physical health problems, and 744 controls.

The response rate was higher among parents than among children (60.5% versus 54.4%). As described earlier, the term 'parents' is generally used because these proxies were most important in the NS-CSHCN-CH. This was again demonstrated by a detailed analysis of the respondents of phase II: mothers completed 85.4% of the questionnaires, fathers completed 13.1%, and both parents together or one parent with his/her new partner completed 0.4%. All other respondent categories (e.g., step

parents, grandparents) were represented with a frequency of < 1%. The most frequently selected predetermined reason expressed by participating parents regarding the non-participation of their children was that 'the child does not want to fill out the questionnaire' (45.1%), followed by 'the child cannot fill out the questionnaire (e.g., because he/she is overextended)' (25.8%) and 'the parents do not want the child to fill out the questionnaire' (7.3%). A further 7.3% of parents did not want to provide a reason for the non-participation of their child.

A comparison between participating and non-participating parents/children is provided in Table 1.3. As shown, non-Swiss children and their parents were less likely to participate than were Swiss children and their parents. Furthermore, parents with a higher level of education were more likely to respond than parents with less education. Lastly, girls were more likely to participate than boys.

Table 1.3: Comparison of participating and non-participating parents and children in phase II

	Total	Participant	Non-participant	df	t / X^2	p
Parents	2,651	N = 1,605 (60.5%)	N = 1,046 (39.5%)			
Study groups						
CSHCN mental health problems: N (%)[a]	879 (33.2)	534 (33.3)	345 (33.0)			
CSHCN physical health problems: N (%)[a]	523 (19.7)	327 (20.4)	196 (18.7)	2	1.372	.503
Controls: N (%)[a]	1,249 (47.1)	744 (46.4)	505 (48.3)			
Mean age, year (SD)[a]	11.44 (1.49)	11.45 (1.51)	11.44 (1.47)	2649	-0.145	.885
Male sex (%)[a]	55.9	55.0	57.2	1	1.192	.275
Swiss Nationality (%)[a]	87.57	92.0	80.7	1	72.758	p<.0005
Highest education, mother[b]						
Low (ISCED 1-2) (%)	9.9	6.6	15.3			
Middle (ISCED 3-4) (%)	63.7	64.4	62.4	2	55.925	p<.0005
High (ISCED 5-6) (%)	26.4	28.9	22.3			
Highest education, father[b]						
Low (ISCED 1-2) (%)	6.6	4.5	10.0			
Middle (ISCED 3-4) (%)	49.7	48.4	51.8	2	38.672	p<.0005
High (ISCED 5-6) (%)	43.7	47.0	38.2			

	Total	Participant	Non-participant	df	t / X^2	p
Children	2,651	N = 1,442 (54.4%)	N = 1,209 (45.6%)			
Study groups						
CSHCN mental health problems: N (%)	879 (33.2)	462 (32.0)	417 (34.5)			
CSHCN physical health problems: N (%)	523 (19.7)	281 (19.5)	242 (20.0)	2	2.528	.283
Controls: N (%)	1,249 (47.1)	699 (48.5)	550 (45.5)			
Mean age, year (SD)	11.44 (1.49)	11.45 (1.51)	11.43 (1.48)	2649	-0.248	.804
Male sex (%)	55.9	53.9	58.2	1	5.039	.025
Swiss nationality[a] (%)	87.57	91.8	82.4	1	52.780	p<.0005

Note: CSHCN: children with special health care needs; *ISCED*: International Standard Classification of Education

[a]All data refer to the targeted children of the parents

[b] The original answers were re-coded into three categories [87]: 1) *low* (mandatory schooling or less), 2) *middle* (vocational training or high school), 3) *high* (technical colleges, upper vocational education, university education)

1.4.1.1.4 Participants

We examined children between the ages of 9 and 14 years living in Switzerland. For the present thesis, only children who fulfilled the following two criteria were included: 1) They had to be classifiable as a CSHCN with a mental health problem, a CSHCN with a physical health problem, or a control; and 2) HRQOL information (proxy- and/or self-rating) had to be available. Altogether, 535 *CSHCN with mental health problems,* 327 *CSHCN with physical health problems,* and 744 *controls* fulfilled these inclusion criteria (see Figure 1.4).

The most frequently mentioned mental health problems among CSHCN were attention deficits (*N*=204), followed by learning difficulties (*N*=131), and conduct problems (*N*=53). All other mental health problems were represented with a frequency of < 5% (e.g., anxiety problems, depressive moods, speech problems, sleeping problems, enuresis, Asperger syndrome, autism, etc.).

CSHCN with physical health problems most frequently had diseases of the respiratory system (*N*=106; e.g., asthma), diseases of the musculoskeletal system and connective tissue (*N*=47; e.g., scoliosis), diseases of the nervous system (*N*=31; e.g., epilepsy) or endocrine, nutritional, and metabolic diseases (20 children; e.g., diabetes). All other categories (e.g., certain infectious and parasitic diseases, neoplasms, diseases of the genitourinary system) were represented with a frequency of < 5%.

1.4.2 Aims

The aims of the present thesis were threefold:

1. To systematically review studies about HRQOL among children with various mental disorders, relative to healthy controls, and to describe the limitations of these studies.
2. To assess the influence of the presence of mental or physical health problems on HRQOL and to analyze the effects of item overlap between mental health problems and HRQOL measurements.
3. To examine parent-child agreement in HRQOL in three health status groups (children with mental health problems, children with physical health problems, and healthy children).

2

Health-related quality of life among children with mental disorders: a systematic review

Quality of Life Research

Dey, M., Landolt, M. A. & Mohler-Kuo, M.

ABSTRACT

Purpose: To systematically review studies about the health-related quality of life (HRQOL) of children with various mental disorders relative to healthy controls and to describe limitations in these studies. ***Methods:*** Relevant articles were searched using different databases, by checking reference lists and contacting experts. We included articles that either compared children with mental disorders to healthy controls/norm values or made such a comparison possible. ***Results:*** Sixteen out of 4,560 articles met the pre-defined inclusion criteria. These studies revealed that the HRQOL of children with various mental disorders is compromised across multiple domains. The largest effect sizes were found for psychosocial and family-related domains and for the total HRQOL score, whereas physical domains generally were less affected. The most important limitations in the existing literature include the lack of study samples drawn from the general population, the failure to use self-ratings, not considering item overlap between measuring HRQOL and assessing for the presence of a particular mental disorder, and not determining whether the children were receiving medication for their mental disorder. ***Conclusions:*** Children with mental disorders experience a considerable reduction in HRQOL across various domains. Research studies that avoid previous limitations are crucial to fill existing knowledge gaps.

INTRODUCTION

The World Health Organization (WHO) [88] claims that *mental disorders* are a neglected field relative to *physical disorders*. To achieve a better balance between the scientific and public attention that mental and physical disorders receive, it is reasonable to use this dualistic distinction. Consequently, in this article, we build upon the frequently used definition of the *'International Classification of Disease and Related Health Problems'* (ICD-10) [1] and apply the thereby-constructed distinction between mental and physical disorders as an analytic framework. According to the ICD-10 definition, mental disorders are the *"existence of a clinically recognisable set of symptoms or behaviours associated in most cases with distress and interference with personal functions* [1]*."* In line with this definition, disorders from Chapter V of the ICD-10 are covered by the term *mental disorders*, whereas all categories from the other chapters are treated as *physical disorders*. Mental disorders in the *'Diagnostic and Statistical Manual of Mental Disorders'* (DSM-IV-TR [89]) are defined as in the ICD-10, and the terms are comparable between the two systems.

One possible way to analyze the impact of a specific disorder is to use the concept of *'health-related quality of life' (HRQOL)*, which can be described as a subjective, multidimensional and dynamic construct that comprises physical, psychological and social functioning [17], thereby going beyond checking for the presence of specific symptoms [19]. HRQOL is, among other things, influenced by the characteristics of a particular disorder, and in children by the stage of the child's development [17]. The term *'quality of life' (QOL)* includes the same dimensions as HRQOL, as well as further dimensions [20]. The concept of QOL is not clearly separated from the HRQOL concept in many publications [19]. For simplicity, we will use the more commonly accepted term HRQOL in this article.

Different authors highlight that most of the HRQOL studies published to date have examined the relationship between *physical disorders* and *HRQOL* [19; 26; 28; 43]. That the relationship between *mental disorders* and *HRQOL* has not received the same degree of scientific attention can be partially explained by the methodical challenge called '*item overlap*', which is bigger for mental (especially in psychosocial HRQOL domains) than for physical disorders [8; 27]. Item overlap exists when the HRQOL items, and the items utilized to assess the presence of a particular disorder are similar in

content [8; 27]. According to Katschnig [8], researchers should control for item overlap during statistical analysis.

Despite the above-mentioned challenge, some investigators have examined the impact of mental disorders on HRQOL. In studies involving *adults*, those with mental disorders consistently report lower HRQOL than healthy controls [54; 90; 91]. In general, *children* have been less frequently considered in HRQOL studies than adults [42]. However, it is important to study children separately, because certain issues are specific for this age group (e.g., the impressive progression of their physical and psychosocial development, greater degree of dependence upon adults, and the different prevalence rates and manifestations of mental disorders) [19; 40; 41].

The aims of this systematic review were twofold: first, to systematically review studies about the HRQOL of children with mental disorders versus healthy controls and second, to identify the limitations of existing articles on this topic, so as to enhance the design of future studies. We failed to find any previous systematic reviews that *concurrently* evaluated HRQOL among children with various mental disorders and met the above-mentioned aims.

METHODS

Data sources and search strategy

A literature search was conducted (up to March 2011) to identify studies that (1) compare the HRQOL of children (ages 0-18 years) with mental disorders versus healthy peers/norm values or (2) provide data that makes such a comparison feasible. The search was conducted in two steps. First, the following databases were searched: DARE, the Cochrane database of systematic reviews, CINAHL, Embase, PsychInfo, PsyIndex, Pubmed, NDLDT and ProQuest. Searches were mainly conducted in English, using the following keywords and Boolean operators: (child* *OR* adolescent* *OR* 'school' *OR* 'p(a)ediatric' *OR* 'youth') AND (psychology* *OR* 'psychic' *OR* psychiatr* *OR* 'mental health' *OR* 'mental disorder' *OR* emotional *OR* behavio(u)ral *OR* developmental *OR* 'mood disorder') AND ('Quality of life' *OR* QOL *OR* well-being). Some additional databases were searched in German (e.g.,

databases with German dissertations). Second, the reference lists of relevant articles and book chapters were consulted for additional materials. Experts in this research field were asked whether they had knowledge of any published or unpublished studies about HRQOL in children with mental disorders.

Study selection

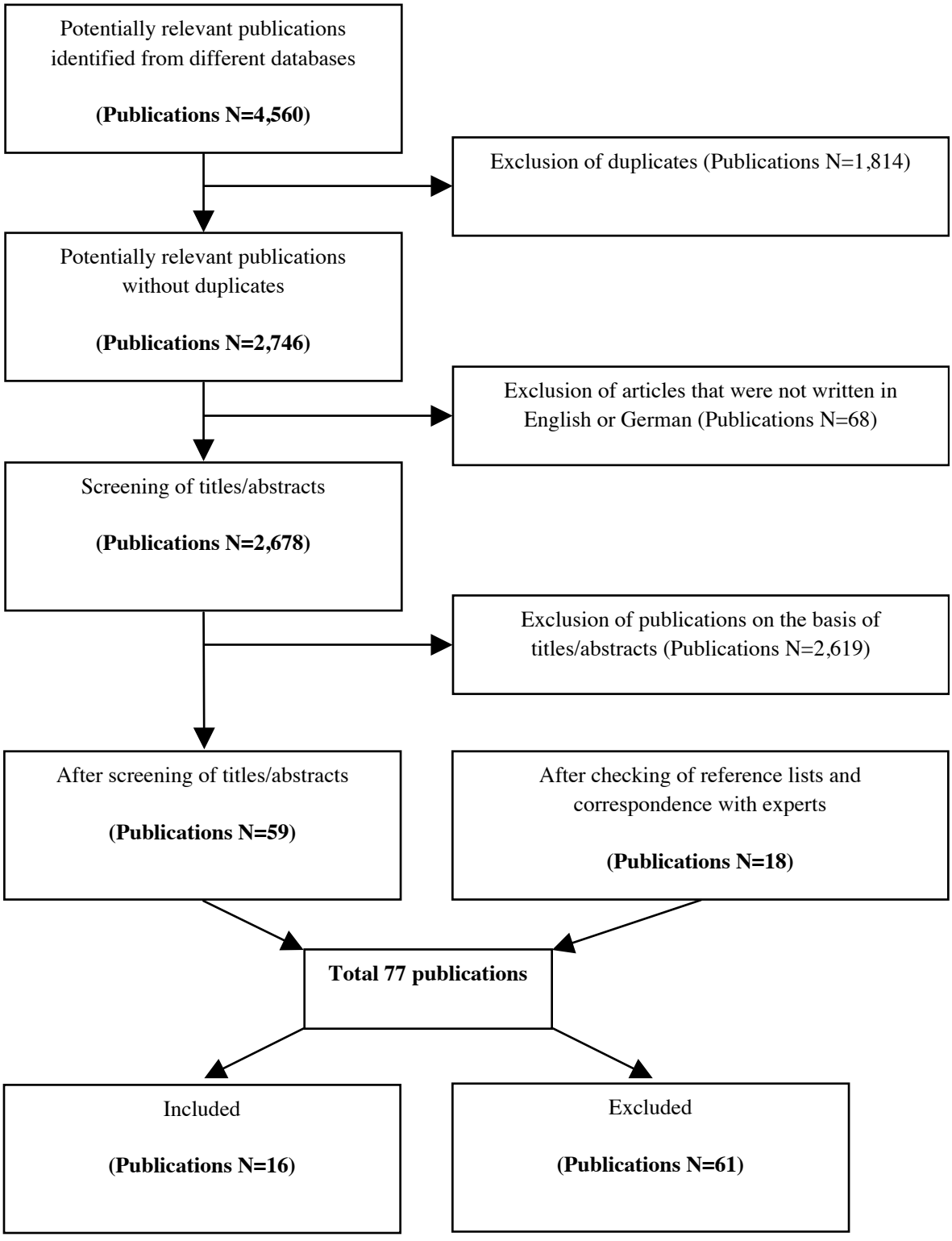
The process of study selection is outlined in Figure 2.1. The first search step revealed 4,560 articles. After eliminating all duplicates (1,814) and those articles not written in English or German (68), 2,678 articles remained. The titles and abstracts of these articles were screened for eligibility by the first author (M.D.). Articles were excluded if at least one of the exclusion criteria was met (see below). Altogether, 2,619 articles were excluded, based upon their title or abstract. The second search step resulted in an additional 18 articles. Full texts of these 18 articles and those articles identified in the databases and not yet excluded (59 articles; for a total of 77 articles) were obtained and reviewed independently by two authors (M.D. and M.A.L.). Papers were excluded if at least one of the following pre-defined criteria was met:

1. Only published as an abstract or poster/no (quantitative) empirical data
2. Data already published in another (included) article
3. Description of mental health and HRQOL of children with physical disorders
4. No disorder from Chapter V of the ICD-10 or DSM-IV-TR
5. Mental disorder diagnosis not confirmed (not diagnosed through a specialist or assessed using a standardized, validated instrument based on ICD or DSM criteria)
6. No standardized HRQOL measure
7. Participants older than 18 years
8. No comparison versus healthy controls/norm values or only a rudimentarily described comparison (if articles did not directly address the differences between children with mental disorders and healthy controls/norms, but provided all the data necessary for this comparison, the article was included)
9. A pharmaceutical study without baseline data

10. More than half of the children with mental disorders were on psychotropic medication during the timeframe to which the HRQOL assessment referred (this criterion was introduced to exclude medical treatment as a potential confounder)
11. Medication unknown and more than half of the children with mental disorders were likely on medication (e.g., children treated in a psychiatric clinic)
12. No descriptive statistics (group means, SD and *N*) reported, computable or provided (to potentially resolve this deficiency, authors were contacted repeatedly and were asked to send us the data)
13. Insufficient quality of reporting (this criterion was applied when multiple concurrent details that normally are reported – like sampling methods, participant details, and statistical analysis methods – were missing).

Inclusion criteria were defined complementary to the exclusion criteria. Disagreements in the appraisal of the articles between M.D. and M.A.L. were resolved through discussion. Ultimately, sixteen publications were included, while 61 were excluded. The reasons for exclusion are described in the Results section.

Figure 2.1: Study selection



Data extraction and synthesis

Two independent reviewers (M.D. and M.M.K.) extracted data from the 16 studies. If crucial information was missing or ambiguous, we asked the authors to send us the missing data or clarify any ambiguity. Concerning study group sizes, we always reported the largest *N* for which HRQOL data were provided. In accordance with Cohen [92], effect sizes (ES) were calculated to evaluate the magnitude of the differences between children with mental disorders and healthy controls/norms. ES also were calculated for studies for which ES were calculated in the reporting paper, because different formulas exist. Each ES was interpreted as *small (0.2)*, *medium (0.5)* or *large (0.8)* in magnitude [92]. *ES ≥ 0.5* were considered *clinically meaningful*. This cut-off was defined according to the recommendation for HRQOL research [93]: It is suggested that a difference of approximately *half a standard deviation (SD)* represents a 'clinically meaningful difference'. Such a difference between the means of children with mental disorders and healthy controls would approximately lead to the here-used cut-off 'ES = 0.5', given the condition that both groups have about the same SD. Furthermore, 95% confidence intervals (CI) were calculated for the ES. Because the included studies differed in relevant characteristics (e.g., specific mental disorders, age range, HRQOL measure), the ES of individual studies were not summarized using meta-analytic methods.

RESULTS

Reasons for exclusion

Reasons for exclusion are listed in Table 2.1. The most common reason for exclusion was the absence or incomplete description of comparisons.

Table 2.1: Reasons for exclusion of articles

Reason for exclusion	**Frequency**
No or only rudimentarily described comparisons	16
More than half of the children with mental disorders were on psychotropic medication	11
Medication unknown and more than half of the children with mental disorders were likely on medication	6
Only abstract or poster / no (quantitative) empirical data	5
Mental disorder diagnosis non-confirmed	5
Data already published in another (included) article	4
Participants older than 18 years	4
No descriptive statistics reported, computable or provided	5
Description of mental health and HRQOL of children with physical disorders (or of a group of children that concurrently included children with mental and physical disorders)	3
No standardized HRQOL measure	1
Insufficient quality of reporting	1

Note: HRQOL: health-related quality of life

Comparing the HRQOL of children with mental disorders versus controls/norms

The 16 studies included in analysis are summarized in Table 2.2. ES are organized by size, with the ES of the total HRQOL score (bold and italic) reported first, followed by the ES of higher-order HRQOL scales (bold) and then the different subscales. ES ≥ 0.5 are underlined because they are considered to be clinically relevant [94]. An overview about the HRQOL measurements that were used in the included studies is provided in Table 2.3.

Table 2.2: Health-related quality of life in children with mental disorders versus healthy controls/norm values (in 16 studies that met final inclusion criteria)

Study	Sample[a]	Age[a]	Comparison (*N*)	Measure	Rater HRQOL	Main outcomes	ES (CI lower limit, CI upper limit) parents	ES (CI lower limit, CI upper limit) children
Attention-deficit/hyperactivity disorder (ADHD)								
Escobar et al. [95]	Clinical	6 to 12	ADHD (120) versus healthy controls (120)	CHQ-PF50	Parent	For most CHQ subscales, children with ADHD had significantly lower scores than healthy children, especially for psychosocial and family-related subscales. In contrast, no significant differences were found in more physical subscales. Both summary scores were significantly lower in children with ADHD than in healthy peers	**PsS: -2.25 (-2.57, -1.92);** **PhS: -0.67 (-0.93, -0.41)** BE: -1.98 (-2.29, -1.67); PE: -1.69 (-1.99, -1.40); FA: -1.42 (-1.70, -1.14); RP: -1.38 (-1.66, -1.10); REB: -1.23 (-1.51, -0.96); MH: -1.23 (-1.50, -0.95); SE: -1.09 (-1.36, -0.82); PT: -0.78 (-1.04, -0.52); FC: -0.53 (-0.79, -0.27); PF: -0.30 (-0.56, -0.05); BP: -0.21 (-0.46, 0.05); GH: -0.18 (-0.43, 0.08)	

Study	Klassen et al. [96]		
Sample[a]	Clinical		
Age[a]	10 to 17		
Comparison (*N*)	ADHD (58) versus norms (parents: 5414; children: 2361)		
Measure	CHQ-PF50 & CHQ-CF87		
Rater HRQOL	Parent & Child		
Main outcomes	ES (CI lower limit, CI upper limit) parents	ES (CI lower limit, CI upper limit) children
Parental rating: Parents of children with ADHD rated the family and psychosocial subscales of HRQOL as substantially reduced, whereas no differences were found in subscales with a stronger relationship to physical health. Child self-rating: Children with ADHD reported reduced HRQOL for only 3 of 9 subscales ('physical function', 'behavior', 'family activities')	BE: -1.85 (-2.11, -1.59); FA: -1.61 (-1.87, -1.34); FC: -1.61 (-1.87, -1.34); SE: -1.05 (-1.31, -0.79); MH: -1.01 (-1.27, -0.75); GH: -0.18 (-0.44, 0.08); BP: -0.03 (-0.29, 0.23); RP: 0.05 (-0.21, 0.31); PF: 0.06 (-0.20, 0.32)	FA: -0.56 (-0.82, -0.30); BE: -0.39 (-0.65, -0.13); PF: -0.37 (-0.63, -0.11); RP: -0.21 (-0.47, 0.05); FC: -0.19 (-0.45, 0.07); MH: -0.04 (-0.30, 0.22); SE: 0.16 (-0.10, 0.42); GH: 0.17 (-0.09, 0.43); BP: 0.20 (-0.06, 0.46)

Matza et al. [97]	Study	
Clinical	Sample[a]	
8 to 17	Age[a]	
ADHD (297) versus norms (391)	Comparison (*N*)	
CHQ-PF50	Measure	
Parent	Rater HRQOL	
Main outcomes	ES (CI lower limit, CI upper limit) parents	ES (CI lower limit, CI upper limit) children
Generally, the CHQ-scores of the ADHD group were reduced for the different psychosocial (sub)scores more than for physical (sub)scales. The baseline mean 'psychosocial summary score' was reduced > 1.5 SD relative to the norm	**PsS: -1.56 (-1.73, -1.39); PhS: 0.70 (0.54, 0.85)** BE: -1.81 (-1.98, -1.63); FA: -1.77 (-1.95, -1.59); PE: -1.72 (-1.90, -1.55); REB: -1.03 (-1.19, -0.87); PT: -1.00 (-1.16, -0.84); SE: -0.83 (-0.99, -0.67); MH: -0.57 (-0.72, -0.41); FC: -0.44 (-0.60, -0.29); RP: 0.02 (-0.13, 0.17); PF: 0.11 (-0.04, 0.27); BP: 0.20 (0.04, 0.35); GH: 0.37 (0.22, 0.52)	

Rentz et al. [98]	Study
Clinical	Sample[a]
6 to 18	Age[a]
ADHD (921) versus norms (391)	Comparison (*N*)
CHQ-PF50	Measure
Parent	Rater HRQOL
Main outcomes	ES (CI lower limit, CI upper limit) parents
Relative to norm values, all psychosocial subscale scores and the 'psychosocial summary score' were significantly reduced in the ADHD group, while the means for the ADHD sample were mostly higher than the norms for physical subscales	**PsS: -1.79 (-1.93, -1.65); PhS: 0.35 (0.23, 0.47)** PE: -1.87 (-2.01, -1.73); FA: -1.67 (-1.81, -1.54); BE: -1.65 (-1.79, -1.52); REB: -1.13 (-1.25, -1.00); SE: -0.99 (-1.11, -0.86); PT: -0.94 (-1.06, -0.81); MH: -0.74 (-0.86, -0.61); FC: -0.50 (-0.62, -0.38); RP: -0.01 (-0.13, 0.11); PF: 0.04 (-0.08, 0.16); BP: 0.06 (-0.06, 0.18); GH: 0.33 (0.21, 0.45)
	ES (CI lower limit, CI upper limit) children

Jafari et al. [100]	Sawyer et al. [99]	Study
Clinical	Non-clinical	Sample[a]
8 to 17	6 to 17	Age[a]
ADHD (72) versus healthy controls (140)	ADHD (308) versus no disorder (2507)	Comparison (*N*)
PedsQL 4.0 generic core scale (23 item)	CHQ-PF50	Measure
Parent & Child	Parent	Rater HRQOL
Parents of children with ADHD and the children with ADHD themselves reported reduced HRQOL values for all (sub)scales and the total HRQOL score	Comparing children with ADHD versus healthy children, large ES were found for the subscales 'behavior', 'parent impact-emotional', 'family activities' and 'parent impact-time'. The smallest ES were identified for subscales with a more physical context	Main outcomes
total: -1.01 (-1.31, -0.71) **PsS: -1.05 (-1.35, -0.75);** **PhS: -0.64 (-0.93, -0.35)** sch: -1.14 (-1.45, -0.84); emo: -0.97 (-1.27, -0.67); soc: -0.42 (-0.71, -0.14)	BE: -1.64 (-1.76, -1.51); PE: -1.30 (-1.42, -1.17); FA: -1.15 (-1.27, -1.02); PT: -0.90 (-1.02, -0.78); REB: -0.77 (-0.89, -0.65); MH: -0.73 (-0.85, -0.61); SE: -0.64 (-0.76, -0.52); GH: -0.29 (-0.41, -0.17); BP: -0.29 (-0.41, -0.18); RP: -0.21 (-0.33, -0.09); PF: -0.19 (-0.31, -0.07)	ES (CI lower limit, CI upper limit) parents
total: -1.12 (-1.42, -0.82) **PsS: -1.09 (-1.39, -0.79);** **PhS: -0.78 (-1.08, -0.49)** sch: -0.95 (-1.25, -0.65); soc: -0.91 (-1.21, -0.61); emo: -0.64 (-0.93, -0.35)		ES (CI lower limit, CI upper limit) children

Preuss et al. [102]	Pongwilairat et al. [101]	Study
Clinical	Clinical	Sample[a]
6 to 18	8 to 12	Age[a]
ADHD (1478) versus norms (1708)	ADHD (46) versus healthy controls (94)	Comparison (*N*)
CHIP-CE	PedsQL 4.0 generic core scale (23 item)	Measure
Parent	Parent & Child	Rater HRQOL
Main outcomes	Parental ratings: The total HRQOL score and all of the psychosocial HRQOL (sub)scales were significantly compromised in children with ADHD versus healthy controls, whereas no differences were found for 'physical health summary score'. Child self-ratings: The total HRQOL score, 'physical health' and all psychosocial HRQOL (sub)scales were significantly reduced in the ADHD group versus controls	According to parent ratings, HRQOL means of the ADHD group were considerably reduced versus a healthy control group for all subscales (ADHD sample scores averaged two SD below the means for healthy controls)
ES (CI lower limit, CI upper limit) parents	***total: -0.73 (-1.09, -0.36)*** **PsS: -0.98 (-1.36, -0.61);** **PhS: -0.26 (-0.61, 0.09)** scho: -1.10 (-1.47, -0.72); emo: -0.67 (-1.03, -0.31); soc: -0.67 (-1.03, -0.30)	ach: -1.92 (-2.00, -1.83); ra: -1.70 (-1.78, -1.62); sat: -1.41 (-1.48, -1.33); res: -1.26 (-1.34, -1.19); com: -0.73 (-0.80, -0.66)
ES (CI lower limit, CI upper limit) children	***total: -0.85 (-1.22, -0.49)*** **PsS: -1.08 (-1.46, -0.71);** **PhS: -0.37 (-0.73, -0.02)** scho: -1.18 (-1.56, -0.80); soc: -0.80 (-1.17, -0.44); emo: -0.71 (-1.07, -0.34)	

Study	Sample[a]	Age[a]	Comparison (*N*)	Measure	Rater HRQOL	Main outcomes	ES (CI lower limit, CI upper limit) parents	ES (CI lower limit, CI upper limit) children
ADHD and additional disorders								
Flapper et al. [103]	Clinical	7 to 10	Development coordination disorder combined with ADHD (23) versus healthy controls (23)	DUX-25; TAC-QOL	Parent & Child	***DUX-25:*** Parental ratings: All HRQOL subscales and the total HRQOL score were significantly lower for the clinical group versus healthy controls. Child self-ratings: Two of the HRQOL subscales ('emotional' and 'social') and the total HRQOL score also were significantly lower ***TACQOL:*** Parental ratings: All but one HRQOL subscale ('bodily functioning') and the total HRQOL score were significantly reduced in ADHD children versus healthy controls. Child self-ratings: All but two HRQOL subscales ('bodily functioning' and 'negative moods') and the total HRQOL score were significantly lower in children with ADHD	**DUX-25**: ***total: -1.06 (-1.68, -0.44)*** home: -1.01 (-1.63, -0.40); phy: -0.97 (-1.58, -0.36); emo: -0.87 (-1.47, -0.26); soc: -0.46 (-1.04, 0.13) **TACQOL**: ***total: -1.52 (-2.18, -0.87)*** SF: -1.78 (-2.46, -1.10); MF: -1.46 (-2.11, -0.81); AF: -1.12 (-1.74, -0.49); NM: -1.11 (-1.73, -0.49); CF: -0.99 (-1.60, -0.38); PM: -0.85 (-1.45, -0.24); BF: -0.31 (-0.89, 0.27)	**DUX-25**: ***total: -1.11 (-1.73, -0.49)*** emo: -1.87 (-2.56, -1.18); soc: -0.76 (-1.36, -0.16); phy: -0.56 (-1.15, 0.03); home: -0.07 (-0.65, 0.51) **TACQOL**: ***total: -1.35 (-1.99, -0.71)*** AF: -1.37 (-2.01, -0.73); SF: -1.33 (-1.97, -0.69); CF: -1.16 (-1.78, -0.53); PM: -0.89 (-1.50, -0.29); NM: -0.74 (-1.34, -0.14); MF: -0.72 (-1.31, -0.12); BF: -0.48 (-1.07, 0.11)

Wehmeier et al. [104]	Study
Clinical	Sample[a]
6 to 17	Age[a]
ADHD with comorbid oppositional defiant or conduct disorder (180) versus norms (14836)	Comparison (*N*)
KINDL-R	Measure
Parent	Rater HRQOL
Main outcomes	ES (CI lower limit, CI upper limit) parents
Compared to published norms for healthy children, the ADHD group had considerably lower HRQOL scores in different domains, with large ES for 5 of 6 subscales and the total HRQOL score. The ES for the 'physical' subscale were very small	***total: -1.13 (-1.27, -0.98)*** fri: -1.21 (-1.36, -1.06); fam: -1.18 (-1.33, -1.04); s-e: -0.92 (-1.06, -0.77); scho: -0.81 (-0.95, -0.66); emo: -0.48 (-0.63, -0.34); phy: 0.06 (-0.09, 0.21)
	ES (CI lower limit, CI upper limit) children

Study	Sample[a]	Age[a]	Comparison (*N*)	Measure	Rater HRQOL	Main outcomes	ES (CI lower limit, CI upper limit) parents	ES (CI lower limit, CI upper limit) children
Conduct disorder								
Sawyer et al. [99]	Non-clinical sample	6 to 17	Conduct disorder (35) versus no disorder (2507)	CHQ-PF50	Parent	In 5 subscales ('behavior', 'family activities', 'parent impact-emotional', 'parent impact-time', 'role/social limitations-emotional/behavioral'), large ES were identified when children with versus children without a conduct disorder were compared. All subscales with a stronger physical component exhibited small ES	BE: -2.28 (-2.62, -1.94); FA: -1.59 (-1.93, -1.26); PE: -1.09 (-1.42, -0.75); PT: -1.08 (-1.42, -0.75); REB: -0.92 (-1.25, -0.58); SE: -0.72 (-1.06, -0.39); MH: -0.62 (-0.96, -0.29); GH: -0.38 (-0.71, -0.04); RP: -0.13 (-0.47, 0.20); BP: -0.12 (-0.45, 0.22); PF: -0.01 (-0.34, 0.33)	

Study	Main outcomes	ES (CI lower limit, CI upper limit) parents	ES (CI lower limit, CI upper limit) children
Specific learning disabilities (SpLD)			

Study	Sample[a]	Age[a]	Comparison (*N*)	Measure	Rater HRQOL	Main outcomes	ES (CI lower limit, CI upper limit) parents	ES (CI lower limit, CI upper limit) children
Rotsika et al. [105]	Clinical	8 to 14	SpLD (99) versus typically developing children (282)	KINDL-R & Kid-KINDL-R / Kiddo-KINDL-R	Parent & Child	Parental ratings: Looking at the descriptive data, HRQOL scores were always lower for the group with SpLD relative to normally developing children (largest ES: 'everyday functioning in school'), except for the 'physical' subscale. Child self-rating: The children with SpLD had lower HRQOL scores for all subscales, compared to normally developing children, with two subscales ('emotional well-being', 'relationship with the family') especially compromised	scho: -1.18 (-1.42, -0.93); s-e: -0.57 (-0.80, -0.34); fam: -0.44 (-0.67, -0.21); emo: -0.34 (-0.57, -0.11); fri: -0.26 (-0.49, -0.03); phy: 0.03 (-0.20, 0.26)	emo: -0.51 (-0.74, -0.28); fam: -0.51 (-0.74, -0.28); scho: -0.44 (-0.67, -0.21); phy: -0.42 (-0.66, -0.19); fri: -0.39 (-0.62, -0.16); s-e: -0.26 (-0.49, -0.03)

Karande et al. [94]				Study
Clinical				Sample[a]
7 to 17				Age[a]
SpLD (150) versus norms (391)				Comparison (*N*)
CHQ-PF50				Measure
Parent				Rater HRQOL
Main outcomes	ES (CI lower limit, CI upper limit) parents		ES (CI lower limit, CI upper limit) children	
The mean subscale and summary scores for children with newly diagnosed SpLD were lower than norm values. Clinically significant ES were discovered for 9 of 12 subscales and the two summary scores	**PsS: -1.33 (-1.54, -1.13); PhS: -1.08 (-1.28, -0.88)** PE: -1.56 (-1.77, -1.35); FA: -1.54 (-1.75, -1.33); PT: -1.36 (-1.57, -1.16); BE: -1.20 (-1.40, -1.00); REB: -1.23 (-1.44, -1.03); GH: -0.96 (-1.16, -0.76); RP: -0.95 (-1.15, -0.75); PF: -0.92 (-1.12, -0.73); MH: -0.71 (-0.91, -0.52); SE: -0.46 (-0.65, -0.27); FC: -0.35 (-0.54, -0.16); BP: -0.34 (-0.53, -0.15)			

Autism spectrum disorder (ASD)	Study					
	Sample[a]					
	Age[a]					
	Comparison (*N*)					
	Measure					
	Rater HRQOL					
	Main outcomes	ES (CI lower limit, CI upper limit) parents		ES (CI lower limit, CI upper limit) children		
	Kuhlthau et al. [106]	Clinical	2 to 17	ASD (286) versus norms (8714)	PedsQL 4.0 generic core scale (23 item)	Parent
	Compared to published norms of healthy children, children with ASD exhibited reduced total HRQOL score and (sub)scale scores (largest ES: 'social functioning'), whereas 'physical functioning' was least compromised	***total: -1.10 (-1.22, -0.98)*** **PsS: -1.39 (-1.51, -1.27); PhS: -0.48 (-0.60, -0.36)** soc: -1.64 (-1.76, -1.52); emo: -0.90 (-1.01, -0.78); scho: -0.74 (-0.85, -0.62)		***total: -0.87 (-1.19, -0.55)*** **PhS: -1.03 (-1.35, -0.71)** soc: -0.76 (-1.09, -0.44); emo: -0.55 (-0.88, -0.23); scho: -0.43 (-0.75, -0.11)		
	Shipman et al. [107]	Clinical	12 to 18	ASD (39) versus norms (parents: 1629; children: 963)	PedsQL 4.0 generic core scale (23 item)	Parent & Child
	Versus published norms, children with ASD and their parents reported significantly lower HRQOL for all domains (children: largest ES: 'physical functioning'; smallest ES: 'school functioning'; parents: largest ES: 'social functioning'; smallest ES: 'physical functioning')	***total: -1.43 (-1.75, -1.11)*** **PhS: -0.71 (-1.03, -0.40)** soc: -1.81 (-2.13, -1.48); emo: -1.24 (-1.56, -0.92); scho: -0.83 (-1.15, -0.51)				

Schizophrenia / schizoaffective disorder						
Study	Sample[a]	Age[a]	Comparison (*N*)	Measure	Rater HRQOL	Main outcomes	ES (CI lower limit, CI upper limit) parents	ES (CI lower limit, CI upper limit) children
Stewart et al. [108]	Clinical	10 to 17	Schizophrenia (10) versus norms (391)	CHQ-PF50	Parent	ES reveal that schizophrenia especially affects psychosocial (sub)scales, whereas physical health is less affected (generally smaller ES). However, some physical (sub)scales still exhibited clinically-relevant ES	**PsS: -3.05 (-3.71, -2.39); PhS: -0.56 (-1.19, 0.07)** REB: -2.92 (-3.58, -2.26); MH: -2.45 (-3.10, -1.80); FA: -2.36 (-3.01, -1.71); PE: -2.30 (-2.95, -1.65); SE: -2.10 (-2.74, -1.45); PT: -2.00 (-2.64, -1.35); BE: -1.86 (-2.50, -1.22); PF: -1.37 (-2.01, -0.74); RP: -0.96 (-1.59, -0.33); FC: -0.38 (-1.01, 0.25); BP: -0.09 (-0.72, 0.54); GH: 0.34 (-0.28, 0.97)	

Stewart et al. [108]	Study
Clinical	Sample[a]
10 to 17	Age[a]
Schizo-affective disorder (7) versus norms (391)	Comparison (*N*)
CHQ-PF50	Measure
Parent	Rater HRQOL

Main outcomes	ES (CI lower limit, CI upper limit) parents	ES (CI lower limit, CI upper limit) children
ES comparing children with schizoaffective disorders versus norm values were especially large for the 'psychosocial summary score' and for related and family-related subscales. In contrast, ES were smaller for the 'physical summary score' and related subscales	**PsS: -3.07 (-3.85, -2.29); PhS: -0.09 (-0.84, 0.66)** FA: -2.94 (-3.72, -2.17); REB: -2.87 (-3.65, -2.10); PE: -2.78 (-3.55, -2.00); BE: -2.22 (-2.99, -1.46); MH: -2.11 (-2.87, -1.35); SE: -1.78 (-2.54, -1.03); PT: -1.60 (-2.36, -0.85); FC: -0.83 (-1.58, -0.08); PF: -0.63 (-1.38, 0.12); BP: -0.46 (-1.21, 0.29); RP: -0.30 (-1.04, 0.45); GH: 0.74 (-0.01, 1.49)	

Study	Sample[a]	Age[a]	Comparison (*N*)	Measure	Rater HRQOL	Main outcomes	ES (CI lower limit, CI upper limit) parents	ES (CI lower limit, CI upper limit) children
Mood disorders								
Freeman et al. [109]	Clinical	8 to 18	Bipolar disorder (89) versus norms (6813)	KINDL-R	Parent	HRQOL (total scale score and all subscale scores) among bipolar children were lower than among healthy controls, especially for psychosocial subscales	***total: -1.96 (-2.17, -1.75)*** fam: -1.70 (-1.92, -1.49); scho: -1.18 (-1.39, -0.97); s-e: -1.33 (-1.54, -1.12); fri: -1.32 (-1.53, -1.10); emo: -1.00 (-1.21, -0.79); phy: -0.55 (-0.76, -0.34)	

Study	Stewart et al. [108]				
Sample[a]	Clinical				
Age[a]	10 to 17				
Comparison (*N*)	Bipolar disorder I (45) versus norms (391)				
Measure	CHQ-PF50				
Rater HRQOL	Parent				
Main outcomes	Comparing bipolar children and norm values, especially large ES are noted for psychosocial and family-related (sub)scales. For the 'physical summary score' and related subscales, the ES were much smaller, but nevertheless sometimes clinically meaningful	ES (CI lower limit, CI upper limit) parents	**PsS: -3.38 (-3.76, -3.00); PhS: -0.04 (-0.35, 0.26)** FA: -3.16 (-3.53, -2.79); MH: -2.72 (-3.07, -2.36); REB: -2.70 (-3.06, -2.34); BE: -2.61 (-2.96, -2.25); PE: -2.41 (-2.75, -2.06); SE: -2.08 (-2.42, -1.74); PT: -2.03 (-2.36, -1.69); FC: -1.15 (-1.46, -0.83); PF: -0.60 (-0.91, -0.29); BP: -0.53 (-0.84, -0.22); RP: -0.39 (-0.70, -0.08); GH: 0.28 (-0.03, 0.58)	ES (CI lower limit, CI upper limit) children	

Study	Sample[a]	Age[a]	Comparison (*N*)	Measure	Rater HRQOL	Main outcomes	ES (CI lower limit, CI upper limit) parents	ES (CI lower limit, CI upper limit) children
Sawyer et al. [99]	Non-clinical	6 to 17	Major depressive disorder (53) versus no disorder (2507)	CHQ-PF50	Parent	Versus healthy children, children with a major depressive disorder exhibited reduced HRQOL in different subscales, with large ES for 'mental health', 'parent impact-emotional', 'role/social limitations-emotional/behavioral', 'family activities' and 'self-esteem'	MH: -1.53 (-1.80, -1.25); PE: -1.32 (-1.60, -1.05); REB: -1.05 (-1.32, -0.78); FA: -0.99 (-1.26, -0.71); SE: -0.83 (-1.11, -0.56); PT: -0.79 (-1.06, -0.51); BE: -0.76 (-1.03, -0.48); BP: -0.72 (-0.99, -0.45); GH: -0.60 (-0.87, -0.32); RP: -0.27 (-0.54, 0); PF: -0.21 (-0.48, 0.06)	

Note: *ADHD*: attention-deficit/hyperactivity disorder; *ASD*: autism spectrum disorders; *SpLD*: specific learning disabilities; *CHIP*: Child Health and Illness Profile; *CHQ*: Child Health Questionnaire; *DUX-25*: Dutch-Child-AZL-TNO-Quality-of-Life; *KINDL-R*: Questionnaire for Measuring Health-Related Quality of Life in Children and Adolescent - Revised Version; *PedsQL*: Pediatric Quality of Life Inventory; *TACQOL*: TNO-AZL-Child-Quality-Of-Life; *HRQOL*: health-related quality of life; *ES*: effect sizes; *CI*: confidence interval; Scales: PsS: psychosocial summary score; PhS: physical summary score; subscales: CHIP: ach: achievement; ra: risk avoidance; sat: satisfaction; res: resilience; com: comfort; CHQ: REB: role/social limitations-emotional/behavioral; BE: behavior; MH: mental health; SE: self-esteem; PE: parent impact-emotional; PT: parent impact-time; FA: family activities; FC: family cohesion; PF: physical functioning; RP: role/social limitations-physical; BP: bodily pain/discomfort; GH: general health perceptions; DUX-25: phy: physical; emo: emotional; soc: social; KINDL-R: fri: friends; fam: family; s-e: self-esteem; scho: school; emo: emotional well-being; phy: physical well-being; PedsQL: sch: school; emo: emotional; soc: social; TACQOL: CF: cognitive functioning; SF: social functioning; MF: motor functioning; AF: autonomic functioning; BF: bodily functioning; NM: negative moods; PM: positive moods

[a] The children with mental disorders

Table 2.3: Overview of the health-related quality of life instruments used in the included studies

Measurement (Abbreviation)[a]/*used version(s)*	***Total HRQOL score*/scales/**subscales (meaning of a positive rated HRQOL)[b]
Child Health and Illness Profile (CHIP) [31] Parent-report: *Child Health and Illness Profile - Child Edition (CHIP-CE) Parent-report form*	Achievement (positive assessment of the way the child performs academically and socially with peers); Risk avoidance (behaviors that pose a risk to one's health/development are avoided); Satisfaction (positive assessment of the child's health and self-esteem); Resilience (positive states and behaviors of the child that are likely to enhance future health); Comfort (no physical and emotional symptoms and limitations)
Child Health Questionnaire (CHQ) [32] Parent-report: *Child Health Questionnaire Parent Form 50 Questions (CHQ-PF50)* Child-report: *Child Health Questionnaire Child Form 87 Questions (CHQ-CF87)*	**Psychosocial Health**[c]; **Physical Health**[d] Role/social limitations-emotional/behavioral (child has no limitations in school work or activities with friends as a result of emotional or behavioral problems); Behavior (child never exhibits aggressive, immature, delinquent behavior); Mental health (child feels peaceful, happy and calm all of the time); Self-esteem (child is very satisfied with abilities, looks, family/peer relationships and live overall); Parent impact-emotional[e] (parent does not experience feelings of emotional worry/concern as a result of child's physical and/or psychosocial health); Parent impact-time[e] (parent does not experience limitations in time available for personal needs due to child's physical and/or psychosocial health); Family activities (the child's health never limits or interrupts family activities nor is a source of family tension); Family cohesion (family's ability to get along is rated 'excellent'); Physical functioning (child performs all types of physical activities, including the most vigorous, without limitations due to health); Role/social limitations-physical (child has no limitations in school work or activities with friends as a result of physical health); Bodily pain/discomfort (child has no pain or limitations due to pain); General health perceptions (child's health is believed to be excellent and will continue to be so)

Measurement (Abbreviation)[a]/*used version(s)*	***Total HRQOL score*/scales/**subscales (meaning of a positive rated HRQOL)[b]
Dutch-Child-AZL-TNO-Quality-of-Life (DUX-25) [33]; adapted from [29] Parent- and child-report: *25 items questionnaire*	***Total HRQOL score*** Home (getting along well with the parents); Physical (positive beliefs/feelings about the physical health; e.g., positive appraisal of his/her power of endurance); Emotional (positive feelings at school, in the night, at this moment); Social (positive feelings about friends and teachers)
Questionnaire for Measuring Health-Related Quality of Life in Children and Adolescent - Revised Version (KINDL-R) [34] Parent-report: *KINDL-R (8-16-years-olds)* Children-report: *Kid-KINDL-R (8-12 years)* *Kiddo-KINDL-R (13-16 years)*	***Total HRQOL score*** Friends (getting along well with peers all the time); Family (getting along well with the parents and feeling fine at home all the time); Self-esteem (feeling well, proud of and pleased with himself/herself and having lots of good ideas all the time); School (enjoying and getting along well in school all the time and never worrying about the future); Emotional well-being (having fun all the time and never feeling listless, alone, scared or unsure of himself/herself); Physical well-being (never feeling ill or low in energy and never having headaches or tummy-aches)
Pediatric Quality of Life Inventory (PedsQL) [35; 36] Parent- and child-report: *PedsQL 4.0 generic core scale (23 items)*	***Total HRQOL score*** **Psychosocial Health Summary Score**[c]; **Physical Health Summary Score**[d] School Functioning (never having problems concentrating, never forgetting things, never having trouble keeping up with schoolwork and never missing school); Emotional Functioning (never feeling anxious, sad, angry, worried and never having any trouble sleeping); Social Functioning (almost always getting along well with peers); Physical Functioning[f] (never having any pain or aches or problems with different physical activities and almost always having a lot of energy)

Measurement (Abbreviation)[a]/*used version(s)*	***Total HRQOL score*/scales/**subscales (meaning of a positive rated HRQOL)[b]
TNO-AZL-Child-Quality-Of-Life (TACQOL) [37-39] Parent-report: *56 item TACQOL PF (parent form)* Child-report: *56 item TACQOL CF (child form)*	Cognitive functioning (never having difficulties with school requirements like paying attention, understanding schoolwork, arithmetic, reading, etc.); Social functioning (never having problems getting along with peers or parents); Motor functioning (never having difficulties with motor functioning - like standing, walking/running, playing, balancing or doing things handily and quickly); Autonomic functioning (never having difficulties doing specific things independently, like going to school on his/her own, going to the lavatory on his/her own, and doing hobbies on his/her own); Bodily functioning (never having physical complaints, like headaches, and never feeling tired, dizzy or nauseated); Negative moods (never having negative feelings, e.g., feeling sad, angry, jealous or anxious); Positive moods (often having positive feelings, e.g., feeling happy, relaxed, enthusiastic or confident)

Note: *HRQOL*: health-related quality of life

Further details about the measurements (e.g., about additional versions) can be found elsewhere (e.g., [19; 22; 26; 28; 29])

[a] only the versions that were used in the included studies (see Table 2.2) are presented in this table, even though some instruments have additional versions

[b] corresponds to the used version (see column 1)

[c] in Table 2.2 called 'psychosocial summary score'

[d] in Table 2.2 called 'physical summary score'

[e] only computable in the parent's version

[f] The 'physical health summary score' contains the same items as the subscale 'physical functioning'. To simplify matters, we therefore only mention the summary score in Table 2.2.

Attention-deficit/hyperactivity disorder (ADHD)

Children with ADHD exhibited reduced HRQOL for multiple parent-rated (sub)scales, with the largest ES identified for psychosocial (e.g., 'behavior', 'parent impact-emotional', 'parent impact-time') and family-related (sub)scales. ES for the parents' ratings usually were smaller for physical (sub)scales. If HRQOL was self-rated, divergent results were evident (in one study, no ES was clinically meaningful; whereas in two other studies, most if not all ES were). Regarding the specific HRQOL domains that were compromised, results similar to those observed with parental ratings were revealed, with the largest ES evident for psychosocial and family-related (sub)scales and smaller ES for most of the physical (sub)scales.

ADHD plus additional disorders

In the study in which ADHD children also had development coordination disorders, the self- and proxy-reports revealed reduced HRQOL in physical, cognitive and social subscales. In another study, the total HRQOL score and different psychosocial subscales of children with ADHD and comorbid oppositional defiant or conduct disorders were reduced.

Conduct disorders

In one study, among children with conduct disorders, all psychosocial (especially for the subscale 'behavior') and family-related HRQOL subscales were clinically meaningfully reduced, whereas no such reduction was apparent in physical subscales.

Specific learning disabilities (SpLD)

The two studies involving children with SpLD identified compromised HRQOL. When parents rated their child's HRQOL, the largest ES were evident in psychosocial (e.g., 'school', 'parent impact-emotional', 'parent impact-time') and family-related (sub)scales. The ES for physical (sub)scales usually were smaller, but sometimes still clinically meaningful. In self-ratings, the ES for children with SpLD were medium for two psychosocial subscales.

Autismus spectrum disorder (ASD)

In two studies, children with ASD had reduced total and subscale scores, both by self- and proxy-report. Parents rated the 'social' subscale as most and 'physical health summary score' least compromised, while children perceived that their physical health was most and 'school' subscale least affected.

Schizophrenia/schizoaffective disorder

Children with either schizophrenia or schizoaffective disorder exhibited reduced HRQOL, with the largest ES identified for psychosocial and family-related (sub)scales. The ES for the 'physical summary score' and related subscales were mostly smaller in magnitude. However, some of these ES were still medium to large.

Mood disorders

Relative to published norms, children with bipolar disorders were reported to have reduced HRQOL, an effect that was again especially pronounced for psychosocial (e.g., 'mental health', 'parent impact-emotional') and family-related (sub)scales. However, the ES were even clinically meaningful for some physical (sub)scales. A similar pattern was identified among children with major depressive disorders.

Limitations of existing studies

Among the *included* studies, the following limitations were apparent and sometimes mentioned by the manuscript authors: First, all but one study [99] used a clinical, rather than a general population, sample. Second, only one study about ASD included children < 6 years old [106]. Third, the majority of studies (62.5%) failed to consider both parental and child HRQOL ratings, reporting only the former. Fourth, the problem of item overlap was addressed in the statistical analyses of one study only [99]. Fifth, even though item overlap sometimes was suggested as a potential explanation, other possible explanations for compromised HRQOL in children with mental disorders were sometimes not provided.

With respect to those articles that were *excluded*, the following two limitations are of special interest (see Table 2.1): First, 17 articles were excluded because more than half of the children with mental disorders were on medication during the time to which the HRQOL assessment referred, or because the medication was unknown and more than half of the children likely were receiving a psychotropic drug. Second, five articles were excluded because the particular mental disorder was not confirmed by a specialist or using a standardized, validated instrument based on ICD or DSM criteria.

DISCUSSION

This systematic review was conducted to compare the HRQOL of children with mental disorders against those of healthy controls/norm values and to describe limitations in the existing literature.

Comparing children with mental disorders versus healthy children/norm values

Parent-ratings

In most of the studies and across various mental disorders, HRQOL was compromised, with ES generally large for total HRQOL scores and psychosocial and family-related (sub)scales, and less (but sometimes still clinically meaningful) for physical (sub)scales.

With regard to psychosocial domains, the largest ES usually were identified among those subscales most closely related to the particular mental disorder (e.g., ADHD and conduct disorders: 'behavior'; SpLD: 'school'; ASD: 'social'; mood disorders: 'mental health'). Some authors considered item overlapping as a possible explanation for this result [97; 99]. Furthermore, it is possible that parents may have over-emphasized the HRQOL aspect that is most closely related to the main problem their child has [105].

In addition, some of the psychosocial subscales not directly associated with the diagnostic criteria of the particular mental disorder were also compromised (e.g., ADHD: large ES in 'self-esteem' [95-98]) – a pattern that possibly emerged due to comorbid disorders [43; 95].

Other subscales that were compromised in various mental disorders describe the impact of the child's mental disorder on the life of the family and parents. This pattern can be explained via different mechanisms; for instance, through parental worries about the present (e.g., meeting daily demands in school) and future (e.g., occupation potential) of their child [105]; and through parental feelings that they are to blame for their child's mental disorder [110]. Furthermore, the impact on parents could be heightened because these children need more support (e.g., doing homework), which leads to less free time for the parents, less time the parents have available for other family members, and their need for greater organizational effort to balance the child's care and parents' work [111].

The clinically meaningful ES for physical (sub)scales that were identified in some studies [94; 95; 97; 99; 100; 103; 107-109] cannot be explained by the side effects of psychiatric drugs [112], because we excluded all studies in which more than half of the children with mental disorders were taking or were assumed to be taking psychiatric medication. However, it is possible that some of the physical (sub)scales were compromised due to comorbid physical disorders [112]. Furthermore, it must be highlighted that some items of the physical subscales had a strong relationship to specific mental disorders. For instance, one item of the 'physical well-being' subscale of the KINDL-R [34] asks whether the child was *tired and worn-out* – something that is also considered a typical symptom for depression.

Looking at the ES of different disorders in Table 2.2, it seems that children with schizophrenia, schizoaffective disorder and bipolar disorder experienced especially compromised HRQOL [108]. However, on closer inspection, what stands out is that the ES differ considerably between studies assessing the same mental disorder. This can be explained through methodological differences. For instance, the way that the participants were sampled seems to influence the magnitude of the ES: When the HRQOL of ADHD children was assessed using the CHQ-PF50 [32], the ES in psychosocial and family-related HRQOL domains were mostly smaller in a study with a non-clinical sample [99] compared to other investigations that used clinical samples [95-98]. This pattern may be explained through the bias that is associated with utilizing clinical samples (see below). Beside the influence of the sampling strategy, other differences between the included studies presumably exerted some influence on the results in general and on the magnitude of the ES in particular. Thus, the differences

between the used HRQOL measurements must be especially emphasized. Even though all of the generic HRQOL measurements that are described in Table 2.3 cover physical, psychological and social HRQOL domains [29], the operationalization of these superordinate domains differ across measures [22; 29]. Hence, when interpreting the results of HRQOL studies, a detailed analysis of the HRQOL measures that are used is necessary. Furthermore, it seems to be easiest to compare the impact of various mental disorders when the methods used (e.g., the sampling protocol and HRQOL measurement) are identical for each mental disorder. This requirement generally is fulfilled in studies that concurrently targeted various mental disorders. Such investigations found that, in terms of overall HRQOL, only a few differences between the distinctive mental disorders emerge, but that each mental disorder is associated with a specific pattern of reduced HRQOL subscales, as described previously [99; 113]. The few differences that were identified in the overall HRQOL between various mental disorders may be attributed to the fact that not only the mental disorders themselves, but also other factors (e.g., symptom severity) exert considerable influence on HRQOL [113].

With regard to all the above-mentioned results, one must consider that the reduced HRQOL in children with mental disorders could also be affected by not yet discussed variables like psychosocial distress in the parents. For instance, it has been demonstrated that parental distress is negatively correlated with all parent-reported HRQOL domains of children with a physical disorder. Furthermore, the relationship between the child's impairment and most of the proxy-reported HRQOL domains was mediated by proxy-distress [114]. Similar relationships are conceivable for proxy-reported HRQOL among children with mental disorders. Consequently, studying such relationships must be considered in subsequent investigations.

Child-ratings

The limited number of studies that incorporated child self-ratings do not allow for clear conclusions regarding HRQOL. However, in some studies, a similar pattern of reduced HRQOL as for parent-ratings was evident, with large ES for total HRQOL score and psychosocial (sub)scales, and smaller ES for more physical (sub)scales. In contrast, other studies revealed HRQOL (sub)scale rankings that

differed between children and parents. For instance, in the study on specific learning disorders, the ES for the self-rated 'school' subscale were not clinically meaningful, whereas parents rated this subscale in such a way as to produce the largest ES [105]. The authors provide multiple explanations for this discrepancy: like parents overemphasizing their child's difficulties in school, children underestimating their target problem to prevent themselves from stressful recognition, and children adjusting to their problem so no further limitations are experienced in the HRQOL subscale that targets academic functioning.

Limitations of existing studies and recommendations for further research

As described in 'Results', the first limitation that was noticed among those studies that were *included* in analysis was that all the studies except [99] used clinical samples. This may lead to biased results, because it is possible that children who have both a mental disorder and reduced HRQOL are more likely to be referred to or treated in a clinic, compared to children with mental disorders without a marked reduction in HRQOL [99]. For example, in a recently published study, referred psychiatric outpatients exhibited lower HRQOL scores than students with equivalent levels of emotional and behavioral problems [115]. Hence, studies that use population-based approaches should be considered to validate the results found among clinical samples. The second limitation was that only one study on ASD included children < 6 years old [106]. This can be explained partially by the fact that the disorders that were the focus of these studies generally are diagnosed after a child reaches that age. However, when a mental disorder occurs earlier and can be diagnosed reliably, HRQOL should be assessed at least with parent-ratings. Third, not all authors used children's self-rating of their HRQOL. Precisely because of the subjectivity of the HRQOL construct, it should – whenever possible – also be self-rated [28]. Admittedly, the cognitive abilities of very young children, and specific characteristics of particular mental disorders (e.g., limited reading ability in children with learning disorders) may hamper such self-ratings [8; 27]. Fourth, the problem of item overlap was addressed in the statistical analyses of only one study [99]. These authors found that, even after controlling for item overlap, similar relationships between mental disorders and HRQOL were observable. Hence, although there may be some item overlap, HRQOL nevertheless provides additional information beyond the

symptoms of mental disorders [15; 19]. All the same, the problem of item overlap warrants further evaluation [19]. Fifth, even though item overlap sometimes was suggested as a potential explanation for reduced HRQOL scores, other possible explanations for compromised HRQOL ratings were provided by only certain authors. Subsequent articles should, therefore, address the mechanisms through which HRQOL ratings become compromised in children with mental disorders in greater detail. Hereby, other influential factors must be taken into account (e.g., the distress of parents when they rate the HRQOL of their child or the severity of the mental disorder).

With respect to those papers that were *excluded*, the first notable limitation was that many studies failed to assess the number of children receiving psychotropic medication that could influence HRQOL [27]. Second, the diagnosis of mental disorder often was not confirmed, investigators relying entirely on parental reports. Some of these studies [116] used population-based samples, which often makes diagnosis confirmation too time- and cost-consuming. However, such a population-based approach has other advantages, as in avoiding the biases that can occur when clinical samples are used. Therefore, depending upon the aims of a particular study, one must evaluate which sampling procedure is most appropriate.

Limitations of our study

The ES presented in Table 2.2 should be interpreted with caution. These values should be treated as approximate values, because some studies used only a small sample size of children with mental disorders. Therefore, 95% CI's obtained from these studies were extremely large. Furthermore, it must be kept in mind that the analyzed studies varied methodologically, thereby reducing their comparability. Studies also used specific inclusion and exclusion criteria that could limit the generalizability of our results. Lastly, we were primarily interested to provide a baseline for the comparison of healthy children and children with mental disorders that were not on psychotropic medication (see exclusion criteria). However, a supplementary systematic review should evaluate the differences between children with mental disorders that are on psychotropic medication from those who are not. By doing so, the inclusion of randomized controlled trials would be most appropriate.

CONCLUSIONS

Our review demonstrates that children with mental disorders experience a considerable reduction in HRQOL across various domains. These effects are not just limited to emotional, social and cognitive dimensions closely related to a specific mental disorder. Hence, reduced HRQOL cannot be attributed exclusively to item overlap. For this reason, HRQOL is a useful construct that can help to expand our knowledge regarding the impact of particular mental disorders and ameliorate clinical (e.g., by better integrating the child's perspective into the treatment plan) and public health practices (e.g., by considering and comparing the HRQOL constraints of different disorders for service planning) [19]. This said our understanding of how mental disorders influence HRQOL among children remains immature and considerable research that avoids some of the limitations of prior attempts is yet needed to fill this knowledge gap.

3

Health-related quality of life among children with mental health problems: a population-based approach

Health and Quality of Life Outcomes

Dey, M., Mohler-Kuo, M. & Landolt, M. A

ABSTRACT

Background: Children with mental health problems have been neglected in health-related quality of life (HRQOL) studies. Therefore, the aims of the current study were 1) to assess the influence of the presence of mental or physical health problems on HRQOL; and 2) to analyze the effects of item overlap between mental health problems and HRQOL measurements. ***Methods:*** Proxy- and self-rated HRQOL (KIDSCREEN-27) of children 9-14 years old was assessed across children with mental health problems (*N*=535), children with physical health problems (*N*=327), and healthy controls (*N*=744). Multiple linear regression analyses were conducted with health status, severity of symptoms, status of medication use, sex and nationality as independent, and HRQOL scores as dependent variables. The effects of item overlap were analyzed by repeating regression analyses while excluding those HRQOL items that contextually overlapped the most frequently-occurring mental health problem (attention deficits). ***Results:*** Severity of symptoms was the strongest predictor of reduced HRQOL. However, all other predictors (except for the status of medication use) also contributed to the prediction of some HRQOL scores. Controlling for item overlap did not meaningfully alter the results. ***Conclusions:*** When children with different health constraints are compared, the severity of their particular health problems should be considered. Furthermore, item overlap seems not to be a major problem when the HRQOL of children with mental health problems is studied. Hence, HRQOL assessments are useful to gather information that goes beyond the clinical symptoms of a health problem. This information can, for instance, be used to improve clinical practice.

INTRODUCTION

'Health-related quality of life' (HRQOL) can be described as a subjective, multidimensional and dynamic construct that comprises physical, psychological and social functioning and that is, among other things, influenced by the health condition of the particular person [17]. To date, more studies have been conducted evaluating the HRQOL of individuals with *physical* than with *mental health conditions* [19]. Additionally, more HRQOL studies have targeted *adults* than *children* [42].

In a recent review article [117], children with *various mental health conditions* were found to exhibit compromised HRQOL relative to *healthy peers*. The largest effect sizes (ES) have been identified for the total HRQOL score and various psychosocial scales, whereas the ES for physical scales generally have been smaller. Parent-ratings of a child's HROQL often were most affected in the psychosocial subscale most closely related to the particular mental health condition. One explanation for this observation is so-called *item overlap*, which is defined as content similarities between HRQOL items and the conceptualization of a particular mental health condition. When such overlap exists, HRQOL and mental health problems are inevitably related [8]. To date, most studies have failed to control for item overlap [117]. However, Sawyer et al. [99] analyzed this issue and observed similar relationships between mental disorders and HRQOL, even after controlling for item overlap. Nevertheless, the effects of this mechanism should be evaluated further [8; 19].

Besides non-consideration of item overlap, the following limitations of previous research were extracted in the above-mentioned review article [117]: first, many authors only used proxy-ratings, even though the subjectivity of HRQOL actually demands self-rating [28] whenever the child's cognitive abilities and particular mental health condition permit [8; 27]. Second, many studies failed to capture whether or not the child was receiving medication for the specific mental health condition, even though it is possible that such treatment affects HRQOL. Furthermore, other potentially-influential variables (e.g., severity of the health condition) were often not included in statistical analyses. Third, existing studies are limited by primarily relying on clinical samples. The utilization of such samples may lead to biased results, because mentally ill children who concurrently have compromised HRQOL may be more frequently referred to or treated in clinics than children with mental health problems lacking HRQOL impairment [99; 115]. Studies that use population-based

samples should be used to verify any results obtained from clinical samples [117]. The few existing studies that have utilized population-based approaches have identified reduced HRQOL (especially within psychosocial (sub)scales) among children with mental health problems [99; 116]. Finally, only a limited number of studies have compared children with mental and physical health conditions with regard to their HRQOL (e.g., [95; 99; 112; 116; 118; 119]). Even though the results of these studies are not completely consistent, Danckaerts et al. [27] summarized various studies and concluded that the overall HRQOL score is reduced to the same extent in children with mental and physical health conditions, whereas psychosocial HRQOL domains are more compromised in children with mental, and physical HRQOL domains in children with physical health problems.

Addressing existing research gaps and the aforementioned limitations, the aims of the present study were two-fold: First, we aimed to assess the influence of mental health problems on proxy- and self-rated HRQOL scores in a population-based sample. For comparison's sake, we also intended to assess the influence of physical health problems on HRQOL. Hereby, other potentially-influencing variables (severity of symptoms, status of medication use) were taken into account. Second, we aimed to examine whether item overlap effects the association between mental health problems and HRQOL.

METHODS

Study procedure

The present study used data from the National Survey of Children with Special Health Care Needs in Switzerland which included a sample of children ages 9-14 from all 26 cantons in Switzerland. We chose children under 15 years, as most health surveys have targeted respondents 15 years old or older. Furthermore, selecting this age group, as opposed to much younger children, allowed us to obtain a HRQOL assessment from both the parents and the children themselves. The protocol was approved by the ethics committee of the Canton of Zurich. A two-stage, population-based sampling method was used to obtain a representative sample (for further details see [69]). The original sample consisted of 16,496 children and their parents.

The main purpose of phase I was to screen the children for *special health care needs* (see measurements). We received screening information about 10,830 children (response rate = 65.7%). Based upon the screening, 1,492 children were classified as *children with special health care needs* (*CSHCN*), 9,294 as children without special health care needs (*healthy controls*) and 44 children were not classifiable due to *missing* data. The latter were excluded from further analyses. Based upon additional information that was gathered during this phase, the CSHCN were subdivided into *CSHCN with mental health problems* (*N*=919), *CSHCN with physical health problems* (*N*=543) and *CSHCN with no classifiable main health problem* (*N*=30). The latter were excluded from further analyses.

The main goal of phase II was to collect information about the self- and proxy-rated HRQOL of *all CSHCN* and a group of *randomly selected healthy controls*. However, not all CSHCN could be re-contacted (if the parents refused to be re-contacted after screening or if the survey material for phase I was returned after the data collection component of phase II was already completed). Altogether, 2,658 HRQOL questionnaires were sent out immediately after screening (881 to CSHCN with mental health problems, 524 to CSHCN with physical health problems, and 1,253 to healthy controls), with seven parent-child pairs excluded because they were no longer contactable (2 CSHCN with mental health problems, 1 CSHCN with a physical health problem, and 4 healthy controls). We received questionnaires back from 60.6% of the parents and/or children (see next section) of the remaining 2,651 parent-child pairs.

In phases I and II, it was emphasized that participation was voluntary. By answering the questions, the parents and/or children provided informed consent.

Sample

We examined children living in Switzerland between the ages of 9 and 14 years. The present study only included children from phase II for which information about HRQOL was provided (self- and/or parent-rating). Altogether, 535 *CSHCN with mental health problems,* 327 *CSHCN with physical health problems* and 744 *healthy controls* were included in the current analysis. The demographic characteristics of the three health status groups are presented in Table 3.1. The most frequently

mentioned mental health problems of CSHCN were attention deficits (*N*=204), learning difficulties (*N*=131) and conduct problems (*N*=53). CSHCN with physical health problems most frequently had diseases of the respiratory system (*N*=106; e.g. asthma), diseases of the musculoskeletal system and connective tissue (*N*=47; e.g. scoliosis) and diseases of the nervous system (*N*=31; e.g. epilepsy).

As demonstrated in Table 3.1, the three health status groups differed in terms of their distribution by sex and nationality. CSHCN with mental or physical health problems further differed with respect to the severity of the main health problem and status of medication use. Thereby, the *(very) low severity* groups had a disproportionate large number of CSHCN with physical health problems relative to CSHCN with mental health problems, whereas the opposite pattern was identified among the *average* to *very high severity* groups. CSHCN with physical health problems were more frequently on medication than CSHCN with mental health problems.

Table 3.1: Demographic characteristics of the three health status groups and health characteristics of children with special health care needs

	CSHCN with mental health problems (N = 535)	CSHCN with physical health problems (N = 327)	Healthy controls (N = 744)	X^2	df	p
Age, years (mean ± SD)	11.39 ± 1.45	11.51 ± 1.54	11.46 ± 1.53	8.281	10	.601
Male sex (%)	65.4	56.0	47.0	42.624	2	p <.001
Swiss Nationality (%)	93.6	94.8	89.5	11.644	2	.003
Severity main health problem						
Very low severity (%)	4.7	13.5				
Low severity (%)	17.6	29.1				
Average severity (%)	48.6	39.8		47.401	4	p <.001
High severity (%)	23.6	12.8				
Very high severity (%)	5.6	4.9				
Medication (% yes)	37.6	65.1		61.791	1	p <.001

Note: CSHCN: children with special health care needs

Measures

- *Special health care needs*: To assess special health care needs, the parent-reported *CSHCN Screener* [71] was used. According to this well-validated and widely-used instrument, a child is classified as *having special health care needs* if he/she presently experiences at least one of five health consequences (e.g., item 1: the need for or use of prescribed medicine; item 5: the need for or use of treatment or counseling for emotional, developmental or behavioral problems) that is due to a health condition which has lasted or is expected to last at least 12 months. If the child did not experience any health consequences, he/she was classified as a *healthy control*.
- *Classification of CSHCN:* After screening for CSHCN, the parents were asked to describe the *main health problem* (open answer format) associated with those special health care needs. The responses were coded according to the *International Classification of Disease and Related Health Problems* (ICD-10 [1]) from which the following two subject groups were created: 1) *CSHCN with mental health problems*, if the main health problem associated with having special health care needs could be assigned to one of the disorders listed in Chapter V ('Mental and behavioral disorders') of the ICD-10; and 2) *CSHCN with physical health problems,* if the main health problem associated with special health care needs could be assigned to Chapter I to IV or VI to XIX of the ICD-10. Altogether, 68 CSHCN could not be assigned to either CSHCN with a mental health problem or physical health problem (e.g., because the parents did not report a specific health problem). These children were assigned to CSHCN with a mental health problem if item 5 of the CSHCN Screener was positive (the need for or use of treatment or counseling for emotional, developmental or behavioral problems) [86]. Accordingly, an additional 38 children were assigned to the group of *CSHCN with a mental health problem*. The remaining 30 were excluded from further analysis.
- *Status of medication use:* According to responses to the first item of the CSHCN screener, the 'status of medication use' was dichotomized as yes / no.
- *Severity of the main health problem:* The parents of CSHCN were asked to rate the severity of their child's main health problem on a five-point scale, ranging from '1' (not at all severe) to '5' (very severe).

- *HRQOL*: The proxy- and self-reported *KIDSCREEN-27* [24] was used to assess HRQOL. This instrument is applicable to children ages 8 to 18 years and has been validated internationally. It contains 5 subscales, namely 'physical well-being', 'psychological well-being', 'autonomy & parent relation', 'social support & peers' and 'school environment'. A 5-point response scale is used for rating, with scores ranging from '1' (not at all/never) to '5' (extremely/always). Because the five subscales differ in the number of items, the sum scores for each subscale were standardized to a scale ranging from 0 to 100, whereby higher scores indicate better HRQOL. Furthermore, in accordance with the manual, a total HRQOL score was calculated that was based on the sum of 10 items. This summation score was also standardized to a scale ranging from 0 to 100. For the overall sample, internal consistency (Cronbach's α [120]) ranged from 0.78 to 0.88 for parental ratings and from 0.74 to 0.83 for child-ratings.

Statistical Analyses

All analyses were conducted using SPSS Version 17.0 for Macintosh [121]. Analyses were performed with two-sided tests and $p<.05$ was considered significant.

Differences in the demographic and health characteristics among the three health status groups (CSHCN with mental health problems, CSHCN with physical health problems, and healthy controls) were compared using χ^2 analyses.

Multiple linear regression analyses were used to assess the association between HRQOL (proxy- and self-rated HRQOL scores) and related predictors, which included health status, severity of the main health problem, and status of medication use. Variance inflation factors (VIF) were used to assess whether multicollinearity between the independent variables existed. A VIF ≥ 10 was interpreted as indicating the presence of multicollinearity [122].

To analyze the effects of *item overlap*, we selected the largest subgroups of CSHCN with mental health problems: children with attention deficits (N=204). We therefore repeated the above-mentioned multiple linear regression analyses for this subgroup 1) without controlling for item overlap; 2) excluding the HRQOL item exhibiting the greatest degree of item overlap with attention deficits

('Have you/has your child been able to pay attention?'); and 3) excluding the two HRQOL items most closely related to attention deficits (additionally excluding the item 'Have you/has your child got on well at school?'). Because 'school environment' was the only subscale that was affected by controlling for item overlap, only the scores of this subscale are reported.

RESULTS

Descriptive HRQOL data by health status group

The means and SD of proxy- and self-rated HRQOL by health status group are presented in Table 3.2. The means were always highest for healthy controls. For the 'physical well-being' subscale, CSHCN with physical health problems had the lowest scores, whereas CSHCN with mental health problems had the lowest scores for all other HRQOL (sub)scales.

Table 3.2: Means and standard deviations for self- and parent-reported KIDSCREEN-27 scores

	CSHCN with mental health problems mean (SD)	CSHCN with physical health mean (SD)	Healthy controls mean (SD)
Parent-report			
Physical well-being	72.19 (16.83)	69.16 (17.40)	78.39 (14.18)
Psychological well-being	73.90 (14.13)	77.75 (13.05)	82.09 (10.35)
Autonomy & parent relation	73.32 (13.67)	75.82 (12.92)	77.93 (12.86)
Social support & peers	64.01 (21.24)	66.01 (20.16)	72.17 (17.03)
School environment	65.49 (18.24)	76.34 (14.95)	78.60 (14.37)
Total HRQOL score	70.43 (12.36)	74.94 (11.21)	79.14 (10.07)
Child-report			
Physical well-being	73.25 (17.00)	71.77 (16.30)	79.41 (14.24)
Psychological well-being	80.45 (14.79)	82.97 (13.34)	85.97 (11.43)
Autonomy & parent relation	77.59 (15.88)	82.06 (13.60)	83.11 (14.03)
Social support & peers	75.35 (22.28)	77.88 (19.75)	82.34 (16.50)
School environment	72.57 (18.64)	79.39 (14.74)	80.63 (15.79)
Total HRQOL score	76.41 (13.37)	80.53 (11.56)	83.13 (11.04)

Note: *CSHCN*: children with special health care needs; *HRQOL*: health-related quality of life

The number of subjects (*N*) varies between the subscale and total HRQOL scores due to missing data. The largest *N* for parent-ratings was 520 for CSHCN with mental health problems, 321 for CSHCN with physical health problems, and 732 for healthy controls. The largest *N* for child-ratings was 454 for CSHCN with mental health problems, 278 for CSHCN with physical health problems and 686 for healthy controls

Multiple linear regression analyses

Besides health status, the severity of the main health problem, and the status of medication use, we also included sex and nationality as independent variables for multiple linear regression analyses, because these variables differed by health status group (see Table 3.1). Multiple linear regression results are presented in Table 3.3. No indicators of multicollinearity were present (all VIF factors ≤ 10).

For all HRQOL scores (except for self-rated 'autonomy & parent relation' and 'school environment'), the most important predictor of reduced HRQOL was the severity of the main health problem. In contrast, use of medication was not associated with any HRQOL scores. All other independent variables were significantly associated with some HRQOL scores. Within the multiple regression models, the presence of mental health problems predicted better parent-reported 'physical well-being', and poorer self-reported 'school environment'. The presence of a physical health problem was significantly associated with better parent-reported 'psychological well-being', 'school environment' and 'total HRQOL'. Female sex predicted reduced proxy- and self-reported 'physical well-being' and 'psychological well-being' and increased scores for self-reported 'social support & peers' and parent-reported 'school environment'. Non-Swiss nationality predicted reduced scores for proxy- and self-rated 'physical well-being' and self-reported 'autonomy & parent relation' subscales.

All regression models were significant at $p<.001$ and accounted for 2.9% to 13.8% of the variance in the parent-reported HRQOL scores, and for 3.3% to 6.9% of the variance in the self-reported HRQOL scores.

Table 3.3: Multiple linear regression analyses on parent- and child-reported health-related quality of life (total health-related quality of life and subscales)

	Physical well-being		Psychological well-being		Autonomy & parent relation		Social support & peers		School environment		Total HRQOL	
	Parent β	Child β	Parent β	Child β	Parent β	Child β	Parent β	Child β	Parent β	Child β	Parent β	Child β
Health status												
Controls (reference)												
CSHCN: mental	.222***	.047	.108	.032	.016	-.096	.042	.026	-.106	-.167**	.042	-.113
CSHCN: physical	.069	-.026	.179***	.084	.063	.022	.042	.032	.138**	-.004	.150**	.018
Severity	-.477***	-.283***	-.456***	-.257***	-.207***	-.091	-.268***	-.227***	-.259***	-.069	-.432***	-.172**
Medication	.005	.011	-.033	-.028	.015	.009	.018	.041	-.021	.040	-.017	.008
Female sex	-.116***	-.102***	-.050*	-.075**	.010	.033	.040	.052*	.102***	.041	-.006	-.039
Non-Swiss nationality	-.057*	-.056*	-.028	-.024	-.037	-.086***	-.008	-.031	.009	.030	-.015	-.031
R^2 adjusted	.111	.069	.119	.048	.029	.033	.048	.035	.141	.046	.138	.061
F	32.855	18.095	36.276	12.889	8.572	8.906	14.052	9.500	43.953	12.145	41.778	15.873
df	6, 1528	6, 1387	6, 1557	6, 1409	6, 1533	6, 1366	6, 1547	6, 1398	6, 1566	6, 1389	6, 1527	6, 1364
p	<.001	<.001	<.001	<.001	<.001	<.001	<.001	<.001	<.001	<.001	<.001	<.001

Note: *CSHCN mental*: children with special health care needs with mental health problems; *CSHCN physical*: children with special health care needs with physical health problems; *HRQOL*: health-related quality of life

Both significant and non significant standardized betas are reported; * = $p \leq .05$; ** = $p \leq .01$ *** = $p \leq .001$; Coding severity: no problem (healthy controls; 0) – very severe (5)

Item overlap

All three sets of analyses (without correction for item overlap; excluding one overlapping item; and excluding two overlapping items) are presented in Table 3.4. No indicators of multicollinearity were identified (all VIF factors ≤ 10). The influence of the variable 'children with attention deficits' on 'school environment' decreased with increasing control for item overlap, as indicated by decreasing standardized betas. However, these changes were only small in magnitude.

Table 3.4: Multiple linear regression analyses on parent- and child-reported 'school environment' with and without controlling for item overlap

	Without correction for item overlap		Excluding one overlapping item		Excluding two overlapping item	
	Parent β	Child β	Parent β	Child β	Parent β	Child β
Health status						
Controls (reference)						
Children with attention deficits	-.190**	-.262***	-.181**	-.221**	-.134*	-.158*
CSHCN: physical health problem	.085	-.108	.054	-.082	.055	-.072
Severity	-.222***	.036	-.172*	.012	-.142*	.017
Medication	.047	.087*	.047	.076	.010	.060
Female sex	.093***	.042	.090***	.054	.105***	.063*
Non-Swiss nationality	.014	.054	.016	.053	.034	.065*
R^2 adjusted	.128	.040	.095	.035	.072	.020
F	31.508	8.818	22.996	7.756	17.262	4.779
df	6, 1246	6, 1115	6, 1246	6, 1115	6, 1246	6, 1115
p	<.001	<.001	<.001	<.001	<.001	<.001

Note: *CSHCN*: children with special health care needs

Both significant and non significant standardized betas are reported; * = $p \leq .05$; ** = $p \leq .01$ *** = $p \leq .001$; Coding severity: no problem (healthy controls; 0) – very severe (5)

DISCUSSION

The purpose of this article was to evaluate the HRQOL of children with mental health problems in-depth. Hereby, some of the research gaps and limitations of existing studies were considered: most important was that 1) a large population-based sample was used; 2) the issue of item overlap was addressed; and 3) in addition to the CSHCN with mental health problems and healthy controls, children with physical health problems were included.

The descriptive statistics of our study are partially consistent with the pattern that was proposed in a review article [27]: That is, the lowest means for *psychosocial HRQOL domains* were identified among children with mental health problems, whereas the lowest means for *physical HRQOL domains* were apparent among children with physical health problems. However, when one only considers the descriptive statistics, it seems that our results contradict the proposition that *overall HRQOL* is equally compromised in children with mental and physical health problems [27]. This being said, the finding of lower total HRQOL scores among CSHCN with mental versus physical health problems was probably due to the severity of the main health problem being greater for the former group. Accordingly, multiple regression analyses indicated that reduced HRQOL was primarily associated with increased severity of the main health problem. This effect was also apparent for total HRQOL.

Despite the clear importance of the severity of the main health problems in predicting HRQOL, mental and physical health problems also contributed to the prediction of some HRQOL scores. For instance, the presence of mental health constraints was significantly associated with a poorer 'school environment' when self-reports were considered, and tended towards poorer 'school environment' when parent-reports were used. The effect on this subscale may be due to the composition of our sample, as the most frequently-reported mental health problems were attention deficits, learning difficulties and conduct problems, thus problems that share their strong impact upon school context. It seems that this impact was not entirely attributable to content similarities between the conceptualization of mental health problems and HRQOL items, because our results remained largely unchanged even when we controlled for item overlap (comparable to findings by Sawyer et al. [99]). Other psychosocial subscales (e.g., 'psychological well-being') were not reduced in CSHCN with

mental health problems in multiple regression models. This might be because this subscale was less directly affected by the most frequently-represented mental health problems in our sample.

That the presence of a mental health problem was associated with enhanced parent-reported 'physical well-being', whereas the presence of a physical health problem predicted higher parent-reported 'psychological well-being' and 'school environment' may be due to compensatory and overly-positive ratings by parents in those HRQOL domains that are not directly related to the particular health constraint [105].

The status of medication use had no influence on HRQOL. This might be due to our inclusion of a variety of different health constraints in our study that were not treated uniformly. Hence, it is possible that the positive effects of some drugs on HRQOL were overlaid by the negative effects of others (e.g., drugs that have severe side effects).

The two included demographic characteristics also exerted an influence on some HRQOL scores in the multiple linear regression models. First and comparable with other studies (e.g., [123]), female sex was associated with reduced 'physical well-being' and 'psychological well-being', as well as with increased 'social support & peers' and 'school environment'. Second, non-Swiss nationality had a significant negative impact upon self- and parent-reported 'physical well-being' as well as on self-reported 'autonomy & parent relation'. This result (especially the former) can be explained by the compromised health status of the non-Swiss population (see, for instance [124]) that leads to reduced HRQOL. However, it also may be due to a more negative assessment of the same health status by non-Swiss relative to Swiss subjects.

Despite the strengths of our study, some limitations must be considered. Similar to other population-based studies (e.g., [116]), the most important limitation was that precise diagnostic information about the main health problem was missing (i.e., CSHCN were classified primarily based upon the parent-reported *main health problem*). However, population-based studies generally uncover similar results as those in which children are diagnosed through a specialist [116]. Furthermore the utilization of clinical samples may lead to biased results [99; 115]. Hence, population-based studies seem to be a worthwhile supplementation to studies with clinical samples [117]. It can be argued that the CSHCN

who were not classifiable based upon their main health problem were classified according to answers to the *fifth item of the CSHCN Screener*. We cannot rule out that the *need for treatment and/or counseling for emotional, developmental or behavioral problems* was the consequence of a significant physical health problem. However, additional analyses indicated that CSHCN with mental health problems who were classified according to the parent-reported main health problem were similar, in terms of their HROQL, as CSHCN with mental health problems classified according to the fifth item of the CSHCN Screener. Hence, we assume that applying this second method was valid.

A second study limitation was that we had no information about comorbid conditions, even though it is possible that these conditions might contribute to the prediction of HROQL.

Third, most of the independent variables (e.g. the health status) were based upon answers provided by the parents. Hence, that the explained variances in multiple linear regression models were larger when HRQOL was rated by parents than by children might be explained through shared-rater method variance.

A fourth limitation is that we used standardized HRQOL scores instead of the Rasch-scaled HRQOL scores proposed by the KIDSCREEN-27 developers [24]. We decided against using Rasch-scaled HRQOL scores because they are difficult to interpret. To deal with this problem, the developers proposed recoding these values into T-scores, a transformation that is based upon norm values. However, only the German-speaking part of Switzerland was included to obtain these norm data, whereas we also drew subjects from the French- and Italian-speaking regions of Switzerland.

CONCLUSIONS

The present paper demonstrates the significant contribution of the *severity* of a child's main health problem to predicting HRQOL, a contribution that has implications for the interpretation of the results of other studies. That is, HRQOL differences between children with mental and physical health problems could be more or less pronounced when the severity of health problems is taken into account. Besides the severity of the main health problem, additional variables were important in

predicting HRQOL. One important finding was that the presence of a mental health problem predicted a poorer 'school environment', a HROQL domain that was most closely related to the most-frequently represented mental health problems (attention deficits, learning difficulties, and conduct problems). This finding seems not to be solely attributable to item overlap between mental health problems and HRQOL items, because our results remained much the same when we controlled for item overlap. Hence, HRQOL assessments are useful when attempting to gather wide-ranging information about CSHCN. This information, which goes beyond the clinical symptoms of mental health problems, can be used in many ways: 1) to expand knowledge about the impact of particular mental health constraints; 2) to improve clinical practices (e.g., by considering compromised HRQOL domains in therapy); and 3) to adapt public health practices (e.g., by considering a broad range of different health conditions and comparing their HRQOL constraints, so as to adequately plan services) [19].

4

Assessing parent-child agreement in health-related quality of life among three health status groups

Social Psychiatry and Psychiatric Epidemiology

Dey, M., Landolt, M. A. & Mohler-Kuo, M.

ABSTRACT

Purpose: To examine parent-child agreement regarding a child's health-related quality of life (HRQOL) among three health status groups. **Methods:** Parent-child agreement was evaluated for three health status groups of a population-based sample: 1) children with mental health problems (*N*=461), 2) children with physical health problems (*N*=281), and 3) healthy controls (*N*=699). The KIDSCREEN-27 was used to assess HRQOL. The children were 9 to 14 years of age. **Results:** Intraclass correlation coefficients were mostly good across all HRQOL scores and health status groups. This relatively high level of agreement was also reflected by the following findings: First, the AGREE group was the largest group in three out of five HRQOL subscales in all health status groups. Second, when disagreement occurred, it was often minor in magnitude. Despite this relatively high level of agreement, the means of self-ratings were significantly higher for all HRQOL scores and health status groups than the means of proxy-ratings. These higher self-ratings were especially pronounced among children with mental health problems in certain HRQOL domains. **Conclusions:** Even though the level of parent-child agreement regarding a child's HRQOL is relatively high, it should be considered that children (especially those with mental health problems) often report better HRQOL than their parents. It is, therefore, highly recommended that both proxy- and self-ratings are used to evaluate a child's HRQOL comprehensively.

INTRODUCTION

While it is well established in the literature that discrepancies exist between proxy- and self-reports about emotional and behavioral problems among children [125-128], parent-child agreement regarding a child's *health-related quality of life (HRQOL)* has less frequently been studied, at least to date.

HRQOL can be described as a subjective, multidimensional and dynamic construct that comprises physical, psychological and social functioning [17]. To account for the subjectivity of this construct, a child's subjective perception should be considered [28]. However, in some instances, proxy-ratings are the only means by which to assess a child's HRQOL (e.g., when the child cannot self-rate his/her HRQOL due to suffering from a particular health condition) [18; 19; 22].

Due to possible discrepancies between the ratings of parents and children, it is important to study 1) whether it is useful to consider both HRQOL ratings because they represent two complementary perspectives [19; 129; 130]; and 2) whether proxy-ratings can be used as a substitute for self-ratings when a child cannot or does not want to self-rate his/her HRQOL [129; 130].

Parent-child agreement can be studied via different methods. To date, Pearson product-moment correlations have been used most frequently [129; 130]. However, correlations may be high even when absolute agreement is low [130]. Therefore, intraclass correlation coefficients (ICCs) should be used instead [131]. Sattoe et al. [132] recently introduced another method. It describes whether parents and children agree in their ratings or whether disagreement in either direction occurs (self-ratings < parent-rating; self-ratings > parent-rating). Furthermore, this method can be used to classify the magnitude of disagreement. Lastly, paired-sample t-tests have been used frequently [129; 130] to assess the degree of difference between the two raters.

In two review articles that mainly included physically ill or healthy children [129; 130] as well as in studies among children with mental health problems [95; 105; 107; 112; 113; 116; 133-135] correlations and/or ICCs have ranged from poor to good.

Despite the relatively low correlation coefficients that have been identified in some studies, it was demonstrated that 43% of parent-child pairs agree regarding a child's HRQOL in one sample of children suffering from physical health problems [132]. For the remaining parent-child pairs,

disagreement in both directions was identified (32% self-rating > proxy-rating; 25% self-rating < proxy-rating). However, this disagreement was mostly relatively small in magnitude.

When the means of self- and proxy-ratings were compared, it was established that parents of physically-ill children [130] as well as of children with mental disorders [95; 105; 107; 112; 113; 116; 133-135] rate most HRQOL domains (significantly) lower than the children themselves. With regard to non-clinical samples, Upton et al. [130] proposed that this pattern is reversed (i.e., self-ratings < parent-ratings).

Even though several studies already have assessed agreement regarding self- and proxy-rated HRQOL, certain gaps remain. First, only a limited number of studies have assessed agreement in children with mental health problems. Second, most HRQOL studies that examined the agreement among multiple informants included either healthy children or children with specific health constraints. How the agreement in HRQOL among children with mental health problems differs from children with physical health constraints *and* from healthy children has not yet been studied comprehensively.

The aim of the present study, therefore, was to examine parent-child agreement regarding a child's HRQOL among three health status groups (children with mental health problems, children with physical health problems, and healthy children) using different methods: 1) ICCs and the method proposed by Sattoe et al. [132] were used to study the level of (dis)agreement; 2) paired sample t-tests and the method of Sattoe et al. [132] were used to evaluate whether self- or proxy-ratings are higher; and 3) across all methods, whether differences by health status groups exist was evaluated.

METHODS

Procedures

We used data from the *National Survey of Children with Special Health Care Needs in Switzerland*. The protocol was approved by the ethics committee of the Canton of Zurich. A two-stage population-based sampling method was used to obtain a representative sample of children ages 9-14 from all 26

Swiss cantons. In the first sampling stage, 258 representative cantons/municipalities were chosen. In the second sampling stage, children ages 9-14 residing in these cantons/municipalities were randomly selected. Details about the two-stage sampling procedure used have been described elsewhere [69]. The cantons and municipalities provided valid demographic information about 16,496 children (last and first name, birth date, sex, address, nationality) and their parents (last and first name).

Children under 15 years old were targeted because other large-scale surveys in Switzerland have included respondents ≥ 15 years old. Furthermore, children ≥ 9 years old were chosen in order to obtain self-reports of HRQOL in addition to primary caretaker's proxy-reports (the terms 'parents' and 'proxies' are used interchangeably in this paper, since 99.4% of the HRQOL questionnaires that were of interest for the present article were filled-out by mothers and/or fathers).

The survey consisted of two phases. The main aims of phases I and II were to *screen* children in order to determine whether they have *special health care needs (children with special health care needs: CSHCN)* and to assess their *HRQOL* (see measurements), respectively. In both phases, it was emphasized that participation was voluntary. By answering the questions, the parents and/or children provided informed consent.

In *phase I*, 10,830 children (response rate = 65.7%) were screened. As a result, 1,492 children were classified as *CSHCN*, 9,294 as children without special health care needs (*controls*) and 44 children were not classifiable due to *missing* data (excluded from further analyses). The 1,492 CSHCN were further subdivided into *CSHCN with mental health problems* (*N*=919), *CSHCN with physical health problems* (*N*=543) and *CSHCN with no classifiable main health problem* (*N*=30; excluded from further analyses).

The main goal of phase II was to collect information about the self- and proxy-rated HRQOL of *all CSHCN*. In addition, a group of *randomly selected controls* was invited to participate in the study as a comparison group (due to budget constraints, not all controls were invited to participate in phase II). However, not all CSHCN could be re-contacted, because 1) the parents refused to participate further in the study after the screening of phase I was completed (*N*=42); or 2) because they did not send the screening questionnaire back in time (phase I), before phase II had ended (*N*=45). Altogether, 2,658

HRQOL questionnaires were sent out immediately after screening (881 to CSHCN with mental health problems, 524 to CSHCN with physical health problems, and 1,253 to controls). Of these, 7 parent-child pairs (2 CSHCN with mental health problems, 1 CSHCN with physical health problems, and 4 controls) were excluded because they could no longer be reached. Of the remaing 2,651 parent-child pairs, 1,606 parents and/or children questionnaires were returned (overall response rate = 60.6%; 60.9% for CSHCN with mental health problems, 62.5% for CSHCN with physical health problems and 59.6% for controls). However, only those children with both parent- and child-reports of HRQOL were included in the analyzed sample (*N*=1,441).

Measurements

The well-validated and widely-used *CSHCN Screener* [71] was applied to assess *special health care needs*. According to this parent-reported measure, a child was classified as *having special health care needs* if the following criteria were met: First, the child presently had to experience at least one of five health consequences (e.g., the need for or use of prescribed medicine). Second, this/these health consequence(s) had to be due to a health condition, which had lasted or was expected to last at least 12 months. If the child did not experience any health consequences, he/she was classified as a *control*. Two methods were used to *classify CSHCN*. The *first method* was based upon the parent-reported *main health problem of CSHCN*, which was coded according to the International Classification of Disease and Related Health Problems (ICD-10 [1]): If the reported main health problem described a disorder from Chapter V (mental and behavioral disorders) of the ICD-10, the child was assigned to *CSHCN with mental health problems*. However, if the main health problem was listed in Chapter I to IV or VI to XIX the child was assigned to *CSHCN with physical health problems*. Altogether, 68 CSHCN could not be assigned to either CSHCN with a mental or physical health problem (e.g., because the parents did not specify the main health problem) with this first method. For these children, a *second method* was applied: If item 5 of the CSHCN Screener was affirmed (the need for or use of treatment or counseling for emotional, developmental or behavioral problems) the child was allocated to *CSHCN with a mental health problem* [86]. Accordingly, an additional 38 children became classifiable. The remaining 30 cases were excluded from further analysis.

The parallel self- and proxy-reported versions of the *KIDSCREEN-27* [24] were used to assess *HRQOL*. This internationally-validated instrument is applicable for children ages 8 to 18 years. Five domains ('physical well-being', 'psychological well-being', 'autonomy & parent relation', 'social support & peers' and 'school environment') and a total HRQOL score (based on 10 items) were calculated. All scores were standardized to a scale ranging from 0 to 100, whereby higher scores indicate better HRQOL. Internal consistency (Cronbach's α [120]) of all health status groups and for both proxy- and self-ratings met or exceeded the threshold of 0.70 that is required for group comparisons [136].

Statistical Analysis

Associations between the three health status groups (CSHCN with mental health problems, CSHCN with physical health problems, and controls) and demographic characteristics were assessed using chi-square tests. The following four methods were applied to evaluate level of agreement: 1) ICCs of absolute agreement [131] were utilized to determine the level of *concordance* between the self- and proxy-ratings; ICCs can be interpreted as poor to fair (≤ 0.40), moderate (0.41 - 0.60), good (0.61 - 0.80) or excellent agreement (0.81 - 1.00) [137]. 2) Paired sample t-tests were used to *compare the means* of the self- and proxy-reported HRQOL scores. 3) *Agreement* and the *direction of disagreement* between the self- and proxy-reports were analyzed further, using the method proposed by Sattoe and colleagues [132]; for all HRQOL scores, the following three *agreement groups* were constructed: A) *AGREE* group: children and parents were assumed to agree when the absolute difference between the self- and proxy-rated HRQOL scores was less than 0.5 SD of the score with the largest variability – this threshold value of 0.5 SD was based upon the definition of clinically-meaningful differences in the HRQOL field [93]; B) *CHILD LOW* group: this disagreement group was defined as when the child's self-report of HRQOL was lower than the proxy-report at a level of at least 0.5 SD; C) *CHILD HIGH* group: this disagreement group was defined as when the child's self-rating of HRQOL was higher than the proxy's report of HRQOL at a level of at least 0.5 SD. Chi-square tests then were used to assess whether the health status groups differed in the distribution of these three agreement groups. 4) To calculate the *magnitude of disagreement* across all HRQOL scores, the CHILD LOW and

CHILD HIGH groups were aggregated into one variable, whereby the direction of disagreement was no longer incorporated. This pooled disagreement then was categorized into *minor* (0.5 - < 1 SD), *intermediate* (1 - < 1.5 SD), *major* (1.5 - < 2 SD) and *substantial* (2 SD <) [132]. Major and substantial disagreements were aggregated in the present article due to their small percentage and similar pattern among the three health status groups. Chi-square tests were used to assess whether the health status groups differed in their magnitude of disagreement.

RESULTS

Sample characteristics

The final analyzed sample consisted of 1,441 children ages 9-14 and living in Switzerland, for which both self- and parent-ratings about HRQOL were available. Of this 1,441, 461 were *CSHCN with mental health problems,* 281 were *CSHCN with physical health problems*, and 699 were *controls*. The mean age (SD) was 11.40 years (1.45) for CSHCN with mental health problems, 11.52 years (1.55) for CSHCN with physical health problems, and 11.45 years (1.52) for controls (χ^2_{10}=5.81; p=.83). The percentage of boys was 65.3%, 54.4% and 46.2%, respectively (χ^2_2=40.26; p<.0005). The percentage of Swiss (vs. non-Swiss) children was 94.1%, 94.3% and 89.3%, respectively (χ^2_2=11.66; p=.003).

Intraclass correlation coefficients and paired-sample t-tests

As reported in Table 4.1, the ICCs of most HRQOL scores were good (exception: the ICC for 'physical well-being' was excellent for CSHCN with physical health problems). Furthermore, children's self-reports of HRQOL were significantly higher than parents' reports of HRQOL within all three health status groups.

Table 4.1: Intraclass correlation coefficients and paired-sample t-tests for the comparison of parent- and child-rated health-related quality of life scores by health status group

	ICC	Means (SD)		Paired sample t-tests		
		Parent-rating	Child-rating	*t*	df	*p*
CSHCN mental						
Physical well-being	0.79	72.54 (16.54)	73.37 (16.96)	-1.225	424	.221
Psychological well-being	0.74	74.03 (13.94)	80.46 (14.89)	-10.266	439	*p*<.0005
Autonomy & parent relation	0.68	73.24 (13.66)	77.40 (15.94)	-5.868	426	*p*<.0005
Social support & peers	0.75	64.66 (21.21)	75.30 (22.07)	-11.463	434	*p*<.0005
School environment	0.76	66.08 (18.14)	72.48 (18.63)	-8.235	436	*p*<.0005
Total HRQOL score	0.74	70.79 (11.94)	76.48 (13.31)	-10.124	418	*p*<.0005
CSHCN physical						
Physical well-being	0.84	69.90 (17.62)	71.86 (16.48)	-2.506	262	.013
Psychological well-being	0.68	77.97 (13.55)	82.90 (13.39)	-6.114	268	*p*<.0005
Autonomy & parent relation	0.62	76.05 (12.97)	82.18 (13.59)	-7.168	267	*p*<.0005
Social support & peers	0.71	66.72 (19.67)	77.95 (19.81)	-9.825	270	*p*<.0005
School environment	0.70	76.23 (15.43)	79.39 (14.80)	-3.588	268	*p*<.0005
Total HRQOL score	0.74	75.19 (11.60)	80.66 (11.60)	-8.302	259	*p*<.0005

	ICC	Means (SD)		Paired sample t-tests		
		Parent-rating	Child-rating	*t*	df	*p*
Controls						
Physical well-being	0.77	78.69 (14.01)	79.66 (14.28)	-2.020	648	.044
Psychological well-being	0.71	82.07 (10.29)	85.94 (11.50)	-9.698	669	*p*<.0005
Autonomy & parent relation	0.67	77.91 (12.74)	83.20 (13.87)	-10.192	647	*p*<.0005
Social support & peers	0.67	72.45 (17.13)	82.61 (15.89)	-15.993	664	*p*<.0005
School environment	0.78	78.69 (14.43)	80.69 (15.80)	-3.985	669	*p*<.0005
Total HRQOL score	0.78	79.18 (10.02)	83.19 (11.05)	-11.309	643	*p*<.0005

Note: *CSHCN mental*: children with special health care needs with mental health problems; *CSHCN physical*: children with special health care needs with physical health problems; *HRQOL*: health-related quality of life; *ICC*: intraclass correlation coefficient

The number of subjects (*N*) varies between the domain and total HRQOL scores due to missing data. The largest *N* consists of 440 parent-child pairs for CSHCN with mental health problems, 271 pairs for CSHCN with physical health problems and 670 pairs for controls. ICCs represent poor to fair (equal or lower than 0.40), moderate (0.41 to 0.60), good (0.61 to 0.80), and excellent agreement (0.81-1.00) [137].

Agreement and direction of disagreement

The distributions of the three *agreement groups* (*CHILD LOW*, *AGREE*, *CHILD HIGH*) by health status group are depicted in Figure 4.1. Across all health status groups, the following pattern emerged: The CHILD LOW group was least common across all HRQOL domains (range: 8.3% - 22.8%). In contrast, the AGREE group was most common for 'physical well-being', 'autonomy & parent relation' and 'school environment' (range: 46% - 57.5%) and the CHILD HIGH group was most common for 'psychological well-being' and 'social support & peers' (range: 43.7% - 55.2%). Chi-square tests revealed that the distribution of the three agreement groups differed significantly by health status group, in terms of total HRQOL score, 'psychological well-being' and 'school environment'. For the total HRQOL score, as well as for 'psychological well-being', the CHILD HIGH group was largest among CSHCN with mental health problems, followed by CSHCN with physical health problems, and subsequently by controls. The reverse pattern was found for the AGREE group. For 'school environment', CSHCN with mental health problems differed from the two other health status groups, by having an especially large CHILD HIGH group and relatively small AGREE and CHILD LOW groups.

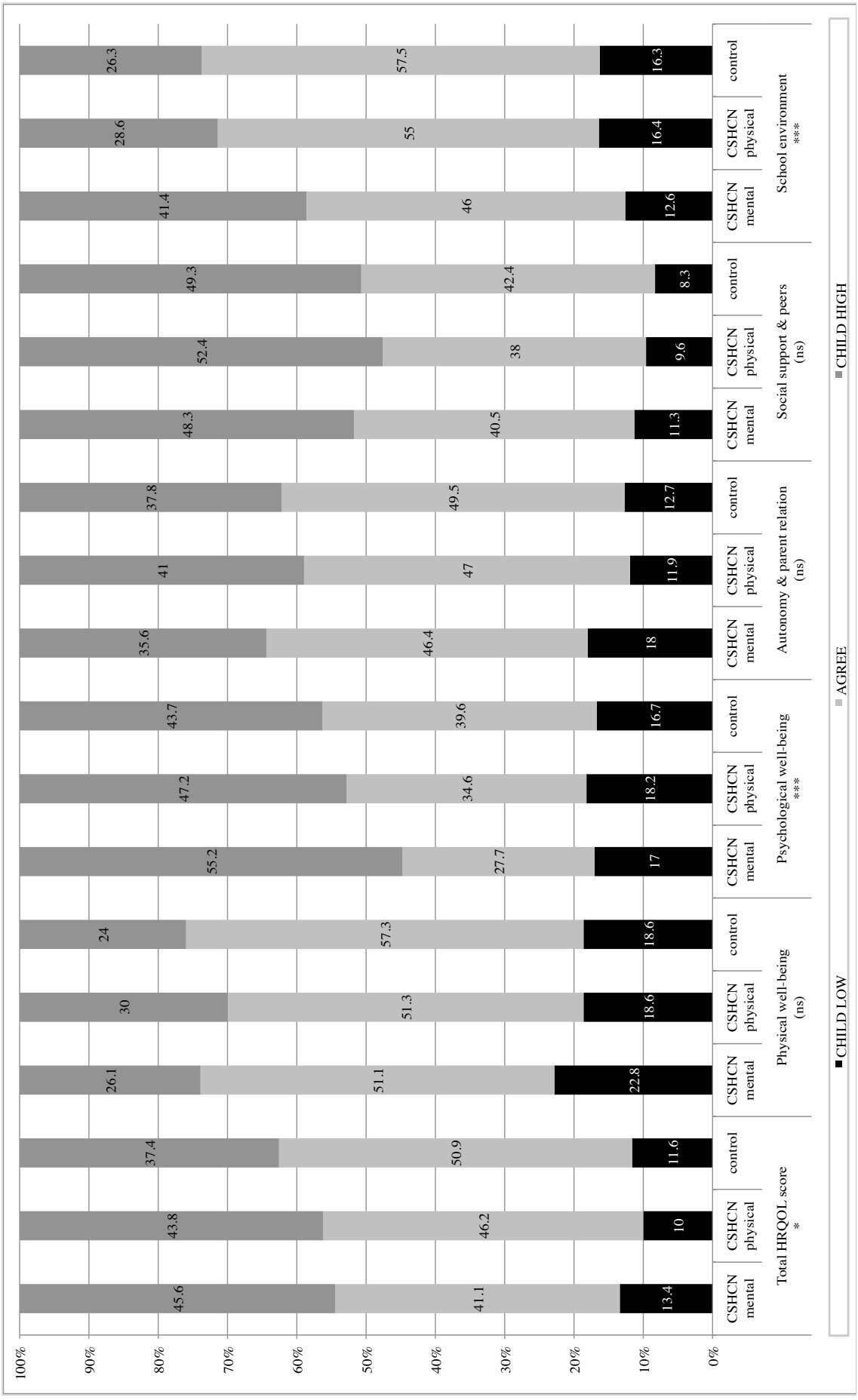


Figure 4.1: Agreement between child- and parent-reports in the KIDSCREEN-27, by health status group

Note: *CSHCN mental:* children with special health care needs with mental health problems; *CSHCN physical*: children with special health care needs with physical health problems; the number of subjects (*N*) varies between the domain and total HRQOL scores due to missing data. The largest *N* consists of 440 parent-child pairs for CSHCN with mental health problems, 271 pairs for CSHCN with physical health problems, and 670 pairs for controls; agreement and direction of disagreement: child – parent score $< \pm 0.5$ (AGREE), ≤ -0.5 (CHILD LOW), ≥ 0.5 (CHILD HIGH) greatest SD of scores (see [132]); Chi-square tests were conducted to evaluate whether a significant association exists between health status group and agreement: ** significant at $p < .01$; *** significant at $p < .001$; ns: not significant

Magnitude of disagreement

The distributions of the *magnitude of disagreement* by health status group are presented in Figure 4.2. Minor disagreement was most common across all health status groups for the domains 'psychological well-being', 'autonomy & parent relation' and 'social support & peers' (range: 47.5% - 64%), whereas major-substantial disagreement was least common across all health status groups for total HRQOL score, 'physical well-being', 'psychological well-being' and 'school environment' (range: 9.1% - 25.5%). On chi-square analysis, the distribution of the magnitude of disagreement differed significantly by health status group for total HRQOL score, 'psychological well-being', 'social support & peers' and 'school environment'. For these, a similar pattern always occurred: the 'minor disagreement' group was relatively small and the 'major-substantial disagreement' group was relatively large for CSHCN with mental health problems, with the reverse pattern identified for controls. Distributions among the CSHCN with physical health problems mostly rested between the two other health status groups.

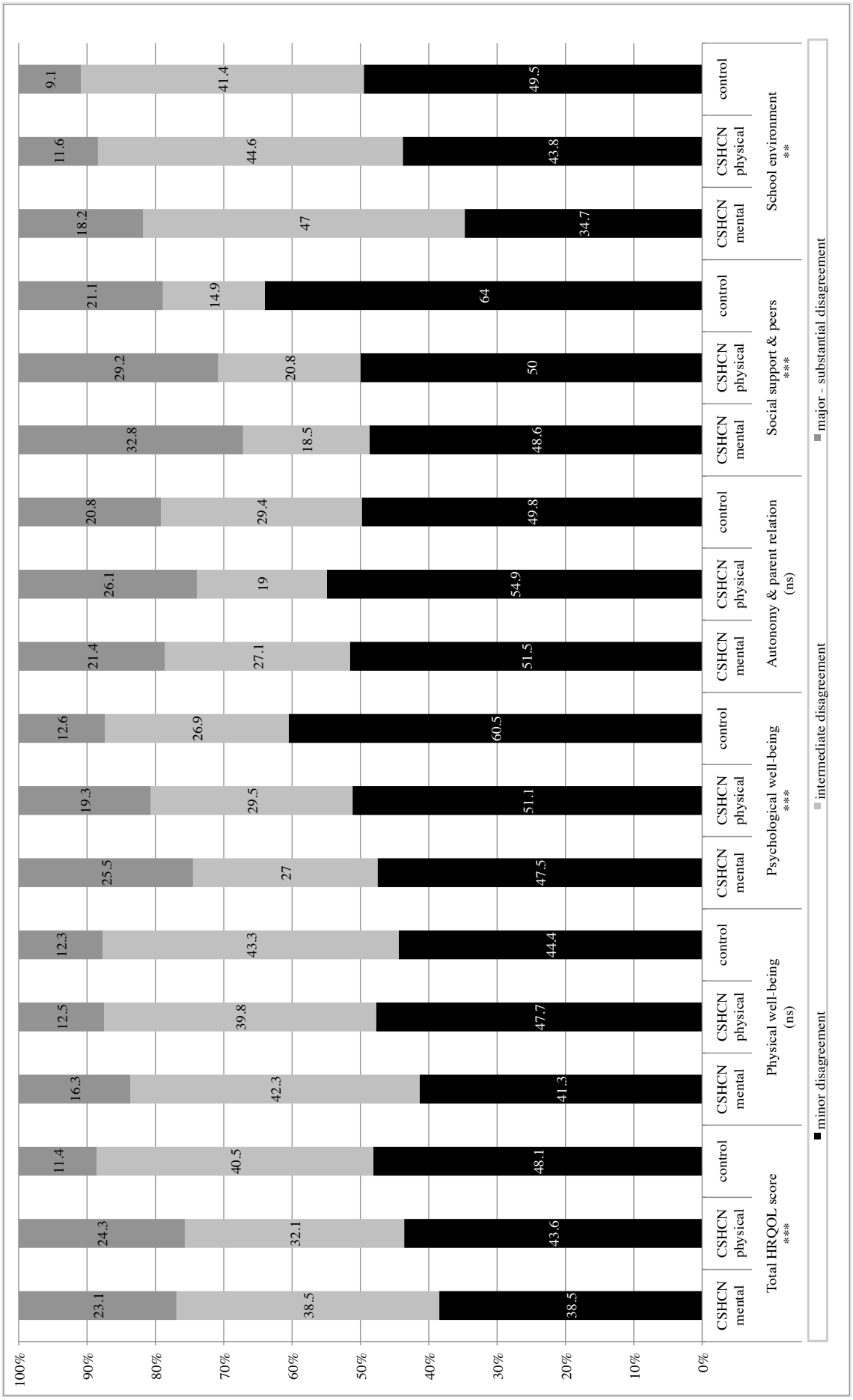


Figure 4.2: Magnitude of disagreement between child- and parent-reports in the KIDSCREEN-27 reports, by health status group

Note: *CSHCN mental*: children with special health care needs with mental health problems; *CSHCN physical*: : children with special health care needs with physical health problems; the number of subjects (*N*) varies between the domain and total HRQOL scores due to missing data. The largest *N* consists of 318 parent-child pairs for CSHCN with mental health problems, 176 pairs for CSHCN with physical health problems and 405 pairs for controls; magnitude of disagreement = child – parent score: *0.5 to < 1* (minor), *1 to < 1.5* (intermediate) *> 1.5* (major - substantial) times the SD of the HRQOL score with the highest variability (see [132]); chi-square tests were conducted to evaluate whether a significant association exists between health status group and magnitude of disagreement: ** significant at $p < .01$; *** significant at $p < .001$; ns: not significant

DISCUSSION

The present study examined levels of parent-child agreement regarding a child's HRQOL in a large, population-based sample of children with different health conditions, using different methods. In all three health status groups, most ICCs were good. This result was consistent with the findings that 1) The AGREE group was the largest group in three out of five HRQOL subscales in all health status groups, and 2) when disagreement occurred, it was often minor in magnitude. Despite this relatively high level of agreement, self-ratings were always significantly higher than proxy-ratings, in all three health status groups. Furthermore, this pattern was especially pronounced among children with mental health problems in some HRQOL domains.

The ICCs that were identified in the current study lay in the upper range of previously-described levels of agreement that ranged from poor to good [95; 105; 107; 112; 113; 116; 129; 130; 133-135]. These relatively high ICCs may be due to any of the following reasons: First, Cronbach's α were sufficient for group comparisons in our study. Hence, the requirement to achieve a high level of agreement was fulfilled [129; 130; 138], whereas it was not met for some domains (e.g., [133]) or subgroups (e.g., [134]) in other studies. This might have decreased ICCs in these studies. Second, the KIDSCREEN-27 [24] has parallel versions for children and parents, whereas the self- and proxy-versions of the HRQOL measurements used in some previous studies were similar, but not identical. Again, this might have reduced agreement in these investigations [130]. Third, in the current study, the HRQOL questionnaires were filled out at home. Hence, it was possible that parents sometimes helped their children to answer the questions, whereby agreement increased.

The relatively high levels of parent-child agreement assessing HRQOL that was established in the current study by ICCs also were confirmed by the results that the group that agreed was often largest across the three health status groups. In addition, when disagreement occurred, it was often minor in magnitude. These findings were further in-line with the results of Sattoe et al. [132]. However, we extended the results of this previous study by demonstrating that the pattern of high agreement or minor disagreement was detectable in different HRQOL domains and across all three health status groups.

Our finding that self-ratings were significantly higher than proxy-ratings among CSHCN with mental and physical health problems is consistent with the results of previous research [95; 105; 107; 112; 113; 116; 130; 133-135]. It is possible that parents rate their child's HRQOL lower due to experienced burdens and concerns associated with the child's health condition [19; 22; 27]. Children, on the other hand, may rate their HRQOL higher, because they do not want to admit how much their health condition affects them [19; 27; 105], because they are not fully aware of restrictions due to this condition [19; 27; 134] or because they have adapted to their situation [8; 105].

However, that higher self-ratings also were identified among healthy controls contradicts the pattern proposed by Upton et al. (self-rating < parent-rating) [130]. On the other hand, our findings were comparable to the results described by Rotsika et al. [105]. Consistently lower proxy- than self-reports may indicate that parents' ratings could be influenced by their concerns and worries or by experienced burdens (e.g., burdens that are due to concurrently having to work and care for their family), or by their own health conditions. Furthermore, it is possible that this general pattern occurred because 1) children tend to provide more extreme answers than their parents; and/or 2) children and parents differ with respect to the reasons they provide for their answers [139] – a pattern that possibly occurs independent of the health status of the child. The higher self- versus proxy-ratings also were confirmed by the method of Sattoe et al. [132] and were in line with this particular study. That is, when children and parents disagreed, the group in which children rated themselves higher than their parents did was always larger than the group in which children rated themselves lower. However, we again extended the findings of Sattoe et al. [132] by illustrating that this pattern emerges regardless of the HRQOL domain or health status group.

The higher self- versus proxy-ratings were especially pronounced among CSHCN with mental health problems within the 'psychological well-being' and 'school environment' domains. Furthermore, CSHCN with mental health problems also were characterized by a relatively large level of disagreement in those HRQOL scores that differed by health status group ('psychological well-being', 'social support & peers' and 'school environment'). That higher self-ratings were especially pronounced in the above-mentioned HRQOL domains may be attributed to the composition of our sample. For example, the most-frequently reported mental health problem was attention deficits. Such

deficits are associated with the above-mentioned HRQOL domains directly (e.g., school functioning) or via comorbid disorders (e.g., mood disorders, which are frequent comorbid disorders among those with attention deficits [140], may influence psychological functioning). Possible reasons for discrepancies between parents and children have been mentioned above. However, it also is possible that they are especially influential in those HRQOL domains that are closely related to a particular health constraint that a child has. Parents of children with attention deficits may, for instance, be particularly burdened and consequently rate their child's HRQOL as especially low in domains that are associated with the school-related problems of their child.

Furthermore, we found that healthy controls have a higher percentage in the AGREE group compared to the other two health status groups. Even when we only examined the parent-child dyads that disagreed (CHILD HIGH and CHILD LOW group), the magnitude of disagreement was smaller in healthy controls compared to the other two health status groups in those HROQL scores that differed by health status groups, especially compared to the group of children with mental health problems. The latter finding is in line with previous studies that described a higher concordance for healthy children and their parents than for children with mental health problems and their caretakers [134].

Despite the strengths of the present study (e.g., assessing parent-child agreement with different methods and among different health status groups), the results should be interpreted within some caution. The most important limitation of this study is that the influence of particular health conditions on agreement could not be studied because 1) detailed diagnostic information about the child's health problem was not available (group composition was based on parent-reports), and 2) some of the mental and physical health constraints that were included rarely occurred. However, it is still meaningful to aggregate different health problems into two categories (CSHCN with mental versus physical health problems), since it can be assumed that children from the same cluster often have very similar challenges. Another study limitation was that the three health status groups differed in their sex and nationality distributions. However, additional analyses revealed similar results for both sexes (boys versus girls) and for the two nationality subgroups (Swiss versus non-Swiss). Hence, it can be assumed that whatever demographic differences existed between our three health status groups likely did not alter our results meaningfully. A last limitation was that the questionnaires were filled out at

home. Hence, the possibility exists that parents helped their children to answer the questions. However, as Varni et al. [116] highlight, such bias would probably be equally distributed across different health status groups.

CONCLUSIONS

Even though the agreement was good to excellent between parents and children with regards to the child's HRQOL, children often reported better HRQOL than their parents. This effect seemed to be especially pronounced among children with mental health problems. Furthermore, the less frequently occurring case scenario that the self-ratings are lower than the proxy-ratings must be considered as well. Due to the various differences, it can be concluded that it is valuable to use both self- and proxy-ratings, because they sometimes represent two different, but equally important perspectives. Furthermore, it must be emphasized that, when proxy-ratings are used as a substitute for self-ratings, the possibility of disagreement must be taken into consideration. This being said, further research clearly is needed to determine which characteristics of the child and/or parents determine whether children and their parents agree or disagree in either direction, in different health status groups and using different statistical methods.

5 General Discussion

5 GENERAL DISCUSSION

The overall objective of the present thesis was to evaluate HRQOL among children with mental health problems, because this group has been sorely neglected in HRQOL studies, to date. The specific aims were: 1) to systematically review publications about the HRQOL of children with various mental disorders relative to healthy controls, and to describe the limitations of these studies; 2) to assess the influence of mental or physical health problems on HRQOL, as well as to analyze the effects of item overlap between mental health constraints and HRQOL measurements; and 3) to examine parent-child agreement regarding a child's HRQOL among three health status groups (children with mental health problems, children with physical health problems, and healthy children). In this final chapter, a summary and overall discussion of results are provided (Section 5.1), followed by a description of the strengths (Section 5.2) and limitations (Section 5.3) of the present study/thesis, suggestions for further research (Section 5.4), and general conclusions (Section 5.5).

5.1 Summary and discussion of results

The main results of the present thesis are summarized and discussed as follows. In Section 5.1.1, the HRQOL of individuals with mental health problems is compared to that of healthy controls. It is also elaborated upon whether it is useful to measure HRQOL among children with mental health constraints, or whether clinical symptoms and HRQOL contents overlap to such an extent that only redundant information can be gathered via diagnostic and HRQOL measurements. In Section 5.1.2, the impacts of mental and physical health constraints on HRQOL are compared. Lastly, parent-child agreement regarding a child's HRQOL is discussed in Section 5.1.3. The identified limitations of existing HRQOL studies about children with mental disorders (see Chapter 2) are addressed throughout all of the following sections (5.1 to 5.4).

5.1.1 HRQOL: Individuals with mental health problems versus healthy controls

5.1.1.1 An Overview

As noted in Section 1.3, *adults* with various mental disorders suffer from reduced HRQOL relative to people with no health constraints. Similar results were identified in our systematic review of the literature (see Chapter 2) that compared the HRQOL of *children* with mental disorders (ADHD, conduct disorder, SpLD, ASD, schizophrenia and schizoaffective disorders, mood disorders) to those of healthy controls or norm values. However, we only included sixteen studies in the systematic review because 1) the number of HRQOL investigations that targeted children with mental disorders is limited; and 2) some of the existing studies had to be excluded because they failed to fulfill our inclusion criteria.

One reason for paper exclusion was that more than half of the children with mental disorders were (assumed to be) on *psychotropic medication* [112; 118; 119; 134; 135; 141-152]. This exclusion criterion was introduced, because it has been demonstrated previously that psychopharmacological therapy might enhance HRQOL among individuals with mental health problems (e.g., [10; 27; 54-57]). However, even these excluded studies confirmed the finding of compromised HRQOL among children with mental health constraints. Hence, pharmacological treatment does not elevate the HRQOL of children with mental health problems to the level of healthy peers. This was possibly because 1) children who are treated with psychopharmacological drugs generally are relatively severe cases; and/or 2) even though medication leads to some HRQOL improvements, it also may be associated with side effects that, in turn, reduce certain HRQOL domains [112]. In the NS-CSHCN-CH, no differences were identified between CSHCN with mental health problems who were on medication and those who were not (see Appendix A.2). Furthermore, the status of medication use had no influence upon HRQOL in multiple regression models (see Chapter 3). However, it is possible that this finding was due to our concurrent inclusion of children suffering from different health constraints who were not treated uniformly. Hence, positive and negative medication effects on HRQOL might have offset each other.

A second reason for excluding publications from our systematic review was that the particular *mental disorder diagnosis was not confirmed* (i.e., not diagnosed by a specialist or assessed using a standardized, validated instrument based upon ICD or DSM criteria; [153]). Such detailed diagnostic information also was missing in the NS-CSHCN-CH. Nonetheless, all of these studies also described how children with mental health problems have compromised HRQOL relative to healthy controls or norm values (the results of the present study are presented in Appendix A.3). That this finding also has been observed in population-based studies (NS-CSHCN-CH; [116; 154]) indicates that the reduced HRQOL among children with mental disorders identified in our systematic review is not solely attributable to the bias that might be associated with clinical samples (such samples were used in 93.8% of the studies analyzed). Rather, it seems that reduced HRQOL occurs not only in children with mental health constraints referred to or treated in a clinic, but also in those who have not yet experienced such a referral or treatment. However, it must nevertheless be considered that the differences between individuals with and without mental health constraints might be greater in clinical compared to population-based samples, due to the above-mentioned source of bias.

In summary, individuals (adults as well as children) with mental health problems have compromised HRQOL relative to healthy controls. In the NS-CSHCN-CH as well as in other empirical investigations, it was demonstrated that this finding is reproducible, even when the children with mental health problems are on medication, when their health problem has not been assessed in detail, and regardless of whether clinical- or population-based samples are used. This being said, even though this result is consistent, it must be considered that it is based on analyses performed at the group level. Hence, it is possible that some individuals with mental health problems rate their HRQOL as satisfactory because they have adapted to their situation and, accordingly, lowered their HRQOL standards [8].

5.1.1.2 Affected HRQOL domains

Having just identified the negative impact of mental health problems on HRQOL in general (see preceding section), the most affected HRQOL domains are now presented. Both in the systematic

literature review (see Chapter 2) and in the present study (see Appendix A.3), children with mental health constraints suffered especially from reduced *psychosocial HRQOL*. Hereby, the strongest negative impact often was identified in those subscales most closely related to the individual's particular mental health condition. For instance, among the reviewed studies, differences between children with ADHD and healthy peers or norm values were particularly pronounced in the HRQOL domains 'behavior' (CHQ; [32]), 'school' (PedsQL; [35; 36]) and 'achievement' (CHIP; [31]). Accordingly, the presence of a mental health problem was the most influential predictor of a poorer 'school environment' in the NS-CSHCN-CH, a HRQOL domain that is strongly associated with the most frequently reported mental health problems (attention deficits, learning difficulties or conduct problems). As demonstrated in Chapter 3, the negative impact on 'school environment' was not entirely attributable to item overlap between the conceptualization of mental health problems and HRQOL items, because our results remained largely unchanged even when we controlled for such contentual similarities.

Furthermore, in the systematic review and the NS-CSHCN-CH (see Appendix A.3), other psychosocial subscales not directly associated with the diagnostic criteria of the particular mental health constraint were compromised, as well (e.g., *'self-esteem'* among children with ADHD [95-98] and *'psychological well-being'* among the here-studied CSHCN with mental health problems, among whom the largest proportion were children with attention deficits). These results might be due to comorbid health problems [43; 95]. For instance, mood disorders that frequently accompany attention deficits [140] might have compromised both of the above-mentioned HRQOL domains (i.e., 'self-esteem' and 'psychological well-being').

Additionally, as reviewed in Chapter 2, children with various mental disorders also have especially compromised HRQOL in domains that describe the *impact* of the child's health constraint upon the life of the *parents and/or family* (a finding that was identified in studies that measured HRQOL with the CHQ [32] or the KINDL-R [34]; see Table 2.2 and 2.3). The KIDSCREEN-27 [24] that was used in the NS-CSHCN-CH also contains a HRQOL domain that assesses family functioning. This domain, which is labeled 'autonomy & parent relation' also was negatively affected by the presence of a mental health problem. However, the impact on this subscale was relatively small compared to those

affecting other HRQOL domains (see Appendix A.3). That the effect on family functioning was less pronounced in our study relative to other investigations may be due to differences in the HRQOL measurements applied. Mental health problems are – among other things – associated with reduced ability of a family to get along (as assessed by the CHQ or KINDL-R), whereas other aspects of family life that are included in the KIDSCREEN-27 may be less affected (e.g., financial resources of the child; the child's availability of free time; the parent's availability of time for the child).

Lastly, in our literature review, *physical HRQOL domains* were generally less affected among children with mental disorders, but sometimes this effect was clinically meaningful. Similarly, the presence of a mental health problem predicted reduced 'physical well-being' in the present study (see Appendix A.3). These findings were not caused by the negative side effects of psychotropic substances, because 1) studies in which more than 50% of the participants were (assumed to be) on medication were excluded from the systematic review; and 2) CSHCN with mental health problems who were on medication reported slightly better 'physical well-being' than those who were not in the NS-CSHCN-CH (see Appendix A.2). However, reduced physical functioning could possibly be explained by physical health problems that were comorbid to the mental health constraints, or by the strong relationship between specific mental health problems and physical functioning. For instance, the KIDSCREEN-27 [24] question of the subscale 'physical well-being' about whether or not the child felt full of energy might also be answered negatively by children who suffer from negative moods. Similar associations are present for other measurements (e.g., for the KINDL-R [34]; see Chapter 2).

In conclusion, mental health constraints compromise psychosocial and parent/family-related HRQOL domains and, sometimes, even physical function. These negative impacts can be explained by the presence of the particular main health problem and by comorbid mental and physical health constraints. However, as explained in Chapters 2 to 4, other possible explanations also must be taken into consideration. The HRQOL ratings of parents of a child with a mental health condition might, for instance, be influenced by the increased burdens imposed by having a sick child (e.g., because they also have to care for the family and concurrently fulfill their work role; [111]), by their feelings of distress [114; 155], by their concerns and worries relating to their child's health issues [19; 22; 27; 105], by their own health problems, or by feeling responsible for the mental health problem their child

has [110]. Self-ratings, on the other hand, also might be compromised in children with mental health problems due to psychopathological fallacies (see Section 1.2.2.2). However, as will be discussed in Section 5.3.2, such fallacies were likely not very influential in the NS-CSHCN-CH.

Furthermore, it was established that item overlap between symptoms of mental health problems and HRQOL items does not (fully) explain the reduced HRQOL among children with mental health problems. That is, controlling for item overlap did not meaningful alter the results, both in the present study as well as in the investigation of Sawyer et al. [99]. Hence, the still existing critique, which claims that HRQOL assessments are tautological because they are too redundant to the diagnostic criteria of mental health constraints, is no longer maintainable. Nevertheless, it has to be acknowledged that – due to the broadness of the HRQOL construct – some contentual similarities between the conceptualization of mental health problems and HRQOL are inevitable. However, similarities between symptoms and HRQOL items are not unique for mental health constraints, but also exist for physical health problems. For instance, the KIDSCREEN-27 item 'Have you been able to run well?' will be answered negatively by children with paraplegia because the inability to run is an inherent component of this physical health problem. In summary, it can be concluded that even though some item overlap exists between symptoms of both mental and physical health problems and HRQOL, HRQOL assessments are still useful for individuals with any health constraints, because additional information can be gathered that completes the clinical picture (also see [15; 19; 30]).

Lastly, it must be emphasized that the negatively-affected HRQOL identified among children with mental health problems might persist into adulthood, because some individuals with childhood-onset disorders still had compromised HRQOL as adults (see Section 1.3). However, the HRQOL of such adults no longer differed from those of healthy controls in other studies (e.g., [156]), though many objective outcomes still indicate some impairments (e.g., regarding the occupational status). This effect might again be attributed to the possibility that people with a long-lasting health condition adapted to their situation and, thereby, lowered their HRQOL standards [8].

5.1.2 Comparing the impact of mental and physical health constraints on HRQOL

Adults with various mental disorders reported comparable or lower HRQOL than individuals with physical health constraints (see Section 1.3). As illustrated in Chapter 3, only a few studies exist that have compared the HRQOL of *children* with mental versus physical health conditions (e.g., [95; 99; 112; 116; 118; 119]). Even though these studies are not completely consistent in their results (probably due to differences in study characteristics), it has been proposed that *overall HRQOL* is reduced to the same extent in children with mental versus physical health conditions, whereas *psychosocial HRQOL* domains are more compromised in children with mental, and *physical HRQOL domains* in children with physical health problems [27].

The results that were identified in the present study at least partly correspond to the proposed pattern: *psychosocial HRQOL domains* were lowest among CSHCN with mental health problems, whereas *physical HRQOL domains* were most compromised among CSHCN with physical health problems (see Table 3.2 and the simple regression analyses in Appendix A.3). However, descriptive statistics as well as simple regression analyses imply that *overall*, mental health problems have a stronger negative effect on HRQOL than physical health problems. As explained in Chapter 3, this conflicting result was probably because CSHCN with mental health problems suffered from more severe conditions than CSHCN with physical health problems. Consistent with this assumption, the severity of the main health problem generally was more influential than the presence of either a mental or physical health problem on multiple regression analysis, an effect that was also apparent for the *total HRQOL* (see Table 3.3).

In summary, both mental *and* physical health conditions are associated with reduced HRQOL. Furthermore, it must be emphasized that the *severity* of the child's main health problem has to be considered when the impacts of mental and physical health conditions on HRQOL are compared. That is, HRQOL differences between children with mental and physical health problems could be more or less pronounced when this predictor is taken into account. It is also possible that not the nature of a particular health problem (i.e., whether it is mental or physical), but its severity is essential for the prediction of HRQOL. A comparison between mental and physical health constraints without controlling for the severity of the particular health problem is nevertheless useful, for comparing the

bio-psychosocial impacts associated with different health conditions and for decision-making relating to the allocation of resources (e.g., for preventive programs).

5.1.3 Parent-child agreement

As reviewed in Section 1.2.1.1, children are usually able to understand HRQOL questions and answer them in a reliable and valid manner when they are 8 years old or older [43; 44]. Consistent with this, the self-reports of the KIDSCREEN-27 [24] can be filled out by children ages 8 to 18 years. However, some children in the present study did not return the HRQOL questionnaire. The most frequently-mentioned reasons for their non-participation were: 1) that they did not want to fill-out the questionnaire and 2) that they lacked the necessary abilities to do so (see Section 1.4.1.1.3). Especially because of these non-participating children, it was important for us to evaluate whether self- and/or proxy-reports about a child's HRQOL must be considered because these two ratings differ [19; 129; 130], and whether proxy-ratings can be used as a substitute for self-ratings when a child cannot or does not want to self-rate his/her HRQOL [129; 130].

Despite the relatively high level of agreement between parents and children that was identified in the present study, it was concluded that both proxy- and self-ratings should be used, whenever possible (also see [19]). This recommendation was formulated because children and parents did not always agree. Self-ratings were often higher than proxy-ratings (for certain psychosocial HRQOL domains, this pattern was especially pronounced among children with mental health constraints); though, for some dyads, self-ratings were lower.

The conclusion that both HRQOL ratings should be used whenever possible also implies that, when a child's HRQOL is only rated by the parents, the rating should not be interpreted as a substitute for the self-rating. Rather, proxy-ratings represent the perspective of parents. Only when it is established in more detail which factors influence such agreement, can proxy-ratings possibly be used to infer how a child would likely have rated his/her HRQOL (see Section 5.4). However, it must be considered that such an inference is based on information that was gained from those parent-child pairs in which the child was willing and able to fill-out the questionnaire. Parent-child agreement might be different

among those dyads in which the child did not want to fill-out the questionnaire or was not able to do so. Hence, estimating self-ratings on the basis of proxy-ratings is always associated with a large degree of uncertainty.

5.2 Study strengths

5.2.1 Strengths of the NS-CSHCN-CH

One of the most important strengths of the NS-CSHCN-CH was its population-based sampling method. For the main aim of the study – to estimate the prevalence of special health care needs – this sampling strategy was more appropriate than, for instance, school-based sampling. The latter would have led to considerable bias, because CSHCN have more school absenteeism than children without special health care needs [157] and, therefore, would have been relatively difficult to reach within a school context.

Another advantage of the applied sampling strategy was that the cantons and municipalities provided basic information about all targeted people. Thus, it was possible for us to assess whether non-response bias existed relating to the socio-demographic characteristics collected (see Appendix A.1; [69; 70]).

Further strengths of the NS-CSHCN-CH included: 1) the large sample size; 2) the inclusion of all Swiss cantons; 3) the utilization of CATI as the method of choice for phase I, which proved to be superior than written questionnaires in various respects (e.g., less missing data, quicker data collection, the ability to clarify ambiguous responses); 4) the concentration on individuals younger than 15 years, because data about this age group have been rare in large-scale Swiss surveys performed to date; 5) the satisfying response rate, which was achieved by different means (e.g., by using mixed methods, see Appendix A.1; [70]); 6) the utilization of a broad definition of special health care needs that led to the inclusion of children who suffered from various and sometimes rare health conditions [67]; 7) asking questions about many different health domains and associated factors (allowing for the processing of various hypotheses and comparisons against data previously collected

within an American survey); and 8) use of the KIDSCREEN-27 [24] to assess HRQOL, given the many advantages of this instrument (see Section 1.4.1.1.3). Furthermore, in the NS-CSHCN-CH and for both proxy- and self-ratings, the internal consistency (Cronbach's α [120]) of all scales and across all health status groups met or exceeded the threshold of 0.70 that is required for group comparisons [136].

5.2.2 Strengths of this Ph.D. thesis

The most important strength of the present thesis was that it focused on *children with mental health problems*, a less-considered age group and clearly-neglected group of health conditions in HRQOL studies up to now (both in investigations that compared HRQOL across different health status groups and in publications on parent-child agreement).

The specific value of the *review article* (see Chapter 2) was that studies on various mental disorders were included, whereby a broad view of HRQOL among children with mental disorders was provided. Furthermore, the calculation of effect sizes was beneficial, because they can be used as approximate values for subsequent comparison studies (provided that similar methods are used). Additionally, it must be highlighted that, to calculate effect sizes, the authors of other papers sometimes had to send us additional material (*N*, means, SD) that was not presented in their own published article(s). Hence, data are sometimes presented here that have not yet been published elsewhere. Lastly, the discussion about the limitations of existing HRQOL studies, as well as the suggestions for subsequent research are useful, because this information will help to improve future studies within this field of research.

With regard to the *empirical part* of this thesis, it was advantageous that not only the relatively frequently studied group of children with attention deficits [27] was included, but also children with less common or recognized mental health problems (e.g., autism). Hence, it was possible to obtain a broader picture about HRQOL among children with mental health problems. Furthermore, it was fruitful that two additional health status groups were considered: healthy controls and children with physical health problems. Lastly, it was valuable that some of the limitations of existing HRQOL studies on children with mental health problems were taken into account during the design of this

study. For instance, self- and proxy-ratings were considered, a population-based sample was used to reduce possible biases associated with clinical samples (see above), and the *N* of the three health status groups was large. With regard to *HRQOL prediction* (see Chapter 3), a further study strength was the consideration of potentially-influencing variables (severity of the main health problem; medication status) and addressing the potentially-problematic issue of item overlap between the diagnostic criteria of mental health constraints and HRQOL items. In terms of the evaluation of *parent-child agreement* (Chapter 4), it was advantageous that we used different statistical methods, each associated with different strengths and limitations [129; 130; 158].

5.3 Study limitations

5.3.1 The lack of detailed diagnostic information

The most important study limitation was that no precise diagnostic information about the *main health problem* of each child was available. As described in Section 1.4.1.1.2, two methods were used to classify CSHCN. The method of choice was to use the parent-reported main health problem (method 1). However, if CSHCN were not classifiable by method 1, they were assigned to 'CSHCN with a mental health problem' if item 5 of the CSHCN Screener was confirmed (method 2).

Method 1 is limited because it is possible that some parents were not knowledgeable enough about their child's main health problem (e.g., because it had not yet been confirmed by a specialist or because the parents were not able to recall the precise label of the diagnosed health problem). Hence, it is possible that HRQOL of the children with mental health problem was better in the present study than it would have been if only children with a diagnosed and possibly more severe mental disorder were included. Nevertheless, several arguments argue in favor of method 1. First, a detailed diagnostic procedure would have been too time- and cost-intensive for the population-based NS-CSHCN-CH. The procedure by which the main health problem was assessed was a compromise to gain such information within a short period of time while not sacrificing the benefits of a population-based study. Second, it can be assumed that most parents were able to provide appropriate information about

their child's main health problem. Other authors [116] have emphasized how population-based studies generally yield similar results as investigations during which children are diagnosed by a specialist.

Classification via method 2 is open to criticism [85; 86] because 1) it was not possible to determine whether the *need for treatment and/or counseling for emotional, developmental or behavioral problems* was due to the main health problem or to the consequences of some physical health condition. For the case scenario in which the physical health constraint was the main health problem and the mental health constraints occurred secondary, the not suitable assignement to CSHCN with mental health problems might have biased the results in such a way that the physical well-being was compromised to a larger and psychosocial HRQOL domains to a smaller extent than for children with a main health problem, which was truly mental in nature; 2) it might have been that some parents mistakenly failed to consider certain health problems as *emotional, developmental or behavioral*; and 3) it is possible that parents did not judge various mental health problems to be sufficiently serious to warrant *treatment or counseling* or were unfamiliar with available resources and services. The two last mentioned scenarios (point 2 and 3) would have led to an exclusion of these children, because they were not classifiable. This exclusion may have led to an overestimated of the negative effects of mental health constraints on HRQOL in the present study, because it can be assumed that children were excluded despite suffering of a mental health constraints only if the severity of their health condition was relatively low. However, the usefulness of adding method 2 is indicated by the following group comparisons. First, when the HRQOL of CSHCN with mental health problems who were classified according to method 1 were compared to the HRQOL of CSHCN classified according to method 2, no significant differences were uncovered (see Appendix A.4). Hence, it seems that the two groups were similar enough to be grouped together. Second, the applied classification was validated through the SDQ ([81; 82]; see Table 1.1 for a description of this measurement): As demonstrated in Appendix A.5, CSHCN with mental health problems had more difficulties in all subscales and in their total difficulties score, relative to CSHCN with physical health problems and healthy controls, whereas fewer results were significant when CSHCN with physical health problems were compared against healthy controls.

In summary, classification of the main health problem via methods 1 and 2 seems to be appropriate within the scope of the NS-CSHCN-CH. Hereby, it also must be emphasized that clinical samples in which individuals generally have a detailed diagnosis might bias results (see Section 5.1.1.1). Therefore, it is advantageous to validate results drawn from clinical samples with those derived via population-based approaches (see Chapter 2).

Besides the lack of detailed diagnostic information about the main health problem, information about *comorbid health conditions* also was limited in the present study (we only asked about the presence of additional health problems that existed besides the main health problem, but not about the nature of these conditions). It is conceivable that such comorbid health constraints also contributed to the prediction of HRQOL (i.e. more comorbid health problems are presumably associated with lower HRQOL). However, it was assumed that the severity of the main health problem, which was included in multiple regression analyses (see Chapter 3), would at least partly mirror the presence of comorbid health constraints. Accordingly, the correlation between the severity of the main health problem and the presence of additional health problems was significant.

5.3.2 Further limitations

One conceptual weakness in the thesis can be seen in the applied fragmentation of *mental* versus *physical health problems*, because mental and physical conditions are closely intertwined [159]. However, this fragmentation was applied because it corresponds to an approach well established both in clinical and scientific settings. Furthermore, the WHO [88] have emphasized how mental health constraints are a neglected field relative to physical health problems. Hence, it can be useful to maintain this dualistic distinction to establish a better balance between the scientific and public attention that mental and physical conditions receive (e.g., when – as in the present thesis – it is revealed that both conditions are associated with reduced HRQOL).

Regarding CSHCN with mental as well as CSHCN with physical health problems, it has to be acknowledged that they were very heterogeneous. Hence, it must be assumed that subgroups of these two health status groups (e.g., children with attention deficits versus children with enuresis) would

also differ in terms of their HRQOL. Regarding different subgroups with mental health problems, it was demonstrated in earlier studies [99; 113], that the *total HRQOL* score is similar across different disorders, whereas differences exist in some *HRQOL domains*. Similar findings are also conceivable for children with different physical health constraints provided that the severity of the health problem is comparable. Therefore, the following can be assumed for the present study: The *total HRQOL* score was not biased by the heterogeneity of the *mental* or *physical health problems cluster* (provided that the various health constraints did not differ to a large extent in regard to their severity). However, the negative effect of a particular health constraint on a HRQOL domain was sometimes weakened by another health constraint of the same mental or physical health problem cluster that did not negatively affect HRQOL (i.e., it is possible that the impact of mental or physical health condition on particular HRQOL domains may have been underestimated in the present study). Furthermore, it is possible that differences between such subgroups would exist with regards to the degree of parent-child agreement about the child's HRQOL. However, due to the scarcity of HRQOL studies that compared the parent and child ratings among different health status groups (see Chapter 4), it is not possible to estimate the bias that might have arised due to grouping heterogeneous groups together. However, for the following reasons, we refrained from comparing these subgroups. On one hand, sometimes, only very superficial information about the main health problem of the child was available (e.g., 'the child suffers from emotional problems'). Hence, the main health problems were not always unequivocally apportionable to specific blocks of the ICD-10. On the other hand, several subgroups about which we had enough information to assign to specific ICD-10 blocks were extremely small (e.g., five children with autism) and therefore less suitable for group comparisons.

Concerning the generalizability of our results, it has to be emphasized that non-Swiss children and/or their parents participated less frequently in the NS-CSHCN-CH than Swiss children and/or their parents – an effect that was at least partly attributable to language/comprehension problems (see Appendix A.1; [70]). Furthermore, other biases must be considered (e.g., that better-educated parents were more likely to participate in phase II than parents with less education). Because a higher socioeconomic status is associated with a better HRQOL (e.g. [160]), it can therefore be assumed, that

HRQOL was overestimated in the present study. However, this bias presumabely occurred equally across all health status groups, since these health status groups did not differ in regard to this variable.

Most limitations relating to the drafted manuscripts (systematic review, HRQOL prediction, parent-child agreement regarding the child's HRQOL) have already been discussed in Chapters 2 to 4. However, regarding the *systematic review* (see Chapter 2) and the *prediction of HRQOL* (see Chapter 3), it can be further argued that – concerning treatment – only the status of medication use was considered, and whether the child was treated otherwise (e.g., psychotherapy) was not ascertained, possibly leading to an over- (if the treatment had a positive effect on HRQOL) or underestimation (if the treatment had no positive effect on HRQOL and if children that are using such treatment have a health constraint of an above-average severity) of HRQOL among children with mental disorders. This was because such forms of treatment have only rarely been studied to date. Hence, it was not reasonable to include this element in our systematic review. Furthermore, we did not gather detailed information about the particular therapy that children received. Conducting any analyses by different therapy forms would not have been possible, given the data collected in the present thesis.

With respect to the article about *predicting HRQOL* (see Chapter 3), the following three limitations also must be considered. First, the effects of item overlap were only discussed for children with attention deficits. Other subgroups were not selected, because it would not have been evident which items should have been excluded (e.g., because the items of the KIDSCREEN-27 [24] have no strong overlap with particular mental health problems) and because some subgroups were too small to investigate these effects thoroughly.

Second, it can be criticized that the three fallacies (see Section 1.2.2.2) that can occur when the HRQOL of individuals with mental health problems are studied were not evaluated. However, such fallacies most likely had at most a small effect in the present study. Children whose main health problem was described as *depressive* or *manic moods* were rarely or not at all represented in the present sample. Hence, any effect of the *affective fallacy* should have been negligible. This being said, it should be noted that depressive moods often occur comorbidly with other mental health constraints (e.g., [43]) and that the affective fallacy might have emerged in such individuals. However, that higher self- versus proxy-ratings were especially pronounced among CSHCN with mental health problems

contradicts this assumption. Furthermore, delusions and hallucinations were never reported among our CSHCN with mental health constraints, such symptoms mostly developing after the age that was studied in the NS-CSHCN-CH [41]. Hence, the *reality distortion fallacy* also should be insignificant in the present study. Lastly, when the child was significantly intellectually impaired, the HRQOL questionnaire generally was only filled out by the parents. Thereby, the risk of bias relating to the *cognitive fallacy* appears minimized.

Third, it must be kept in mind that the NS-CSHCN-CH was a cross-sectional study. Hence, no conclusions about the direction of influence can be made, and it can be assumed that the influences are multidirectional (e.g., the presence of a mental health problem could negatively affect HRQOL, and reduced HRQOL could further reinforce a mental health problem).

Regarding *parent-child agreement* (see Chapter 4), it must be noted that the main health issue of CSHCN with mental health problems was rated more severe than those of CSHCN with physical health problems (see Chapter 3). It is possible, then, that the significantly higher self- relative to proxy-ratings among CSHCN with mental health problems, versus other health status groups, were influenced by this group difference. However, even when health status groups were stratified according to the severity of the main health problem of the child, the pattern of higher self- versus proxy-ratings among CSHCN with mental health problems was largely replicated for 'psychological well-being', 'school environment' and 'social support & peers'.

5.4 Implications for future research

Subsequent studies could be improved by considering the following issues. First, population-based sampling should be combined with some detailed diagnostic procedure to identify the main as well as comorbid health problems. Provided that the population studied is large enough, a detailed diagnostic procedure would also allow for comparing children with various mental health problems, a comparison that, to date, has only rarely been performed (e.g., [99; 113]). Such studies are important because – as reviewed in Chapter 2 – existing HRQOL studies are limited in their comparability, due

to methodological differences. Second, further studies are still needed that compare the HRQOL of children with mental health constraints versus those with physical health problems. Hereby, it is important that the severity of the compared health conditions is considered. Third, it is essential to specifically focus on non-Swiss children in subsequent studies, because they were underrepresented in the NS-CSHCN-CH sample. Fourth, the methodological challenges (item overlapping and psychopathological fallacies) that arise when the HRQOL of individuals with mental health constraints are studied should be (further) evaluated. Fifth, longitudinal studies are required to evaluate the direction of influences on HRQOL. Sixth, not only the effects of medication on HRQOL among CSHCN should be evaluated, but also the effects of other forms of treatment (e.g., psychotherapy). Seventh, more studies are needed on the levels and directions of parent-child agreement about a child's HRQOL (e.g., qualitative research about the reasons parents and children differ in their ratings; see [139]). Lastly, there remains a need for HRQOL studies on very young children who suffer from mental illness.

Besides the suggestions for improving subsequent studies, additional analyzes should be conducted on the basis of available HRQOL data. First, it is important to further evaluate which additional variables (e.g., age, socioeconomic status, mental and physical health of the parents), besides the ones considered in Chapter 3, predict HRQOL in children. Second, predictors of parent-child agreement (e.g., sex and age of the child and health status of the parents) also should be investigated, because so few studies have been conducted on this so far (for an overview: [129; 130]). Third, qualitative analyses of comments by children and parents on the HRQOL questionnaires should be carried out (compare [21]). Such analyses could – among other things – help to understand how a child deals with his/her health condition, thereby complementing the picture attained through quantitative analyses. Exploratory analyses have already been conducted (see Appendix A.6). Lastly, it could be fruitful to classify individuals according to their HRQOL and not according to their health condition [99; 161]. Doing so would potentially account for the observation that individual patients who suffer from the same health condition are not uniform in rating their HRQOL [162].

Finally, it also will be important to update the systematic review of the literature we conducted about HRQOL among children with mental health problems at regular intervals. Once enough high-quality

and methodologically similar studies on this research topic have been published, meta-analysis methods should be applied.

5.5 Conclusions

The present thesis revealed three major observations: *First, children with mental health problems were found to exhibit compromised HRQOL relative to healthy controls, an impact that was especially pronounced in psychosocial and – depending upon the HRQOL measurement utilized – parent- and family-related HRQOL domains*. This pattern seemed not to be caused (solely) by item overlap between the diagnostic criteria of given mental health constraints and HRQOL items. Rather, a HRQOL assessment provides information that goes beyond the symptoms of a mental health condition, thereby helping to complete the picture about the impact that a given condition and its treatment have on individuals. Such a broad bio-psychosocial perspective can, for instance, be used to enhance clinical practice (e.g., by better integrating the child's perspective into the treatment plan; see [19]).

Second, both mental and physical health conditions were found to be associated with reduced HRQOL. Consequently, the current dearth of HRQOL research among those with mental health problems clearly is not justified. Furthermore, this finding could, in the long term, lead to the elimination of current imbalances in resource allocated to mental versus physical health services [19]. We also note that the effect of the severity of a child's main health problem on HRQOL must be considered when mentally and physically-impaired groups are compared, because this is a highly relevant predictor of reduced HRQOL.

Third, despite the relatively high level of parent-child agreement regarding a child's HRQOL, in many dyads, self-ratings were higher than proxy-ratings (a pattern that was especially pronounced among children with mental disorders in some psychosocial domains), though the reverse pattern sometimes occurred as well (self-rating < proxy-rating). Due to this potential for discrepancies, it is recommended that: 1) self- and proxy-ratings should both be used, whenever possible; and 2) when

only proxy-ratings are obtainable, these ratings should not be interpreted as a substitute for self-ratings, but merely as the perspective of the parents, which may be influenced by several factors.

Finally, it must be emphasized that research into HRQOL among children with mental health constraints is still a relatively new field. There is dire need for further studies to fill numerous existing knowledge gaps.

6 References

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APPENDIX

A.1 An analysis of non-response in a Swiss national survey

International Journal of Public Health

Dey, M. & Mohler-Kuo, M.

INTRODUCTION

Survey response rates generally have been decreasing over recent decades [1-4]. For example, response rates for the annual *U.S. National Health Interview Survey* declined from 80.4% in 1997 to 72.5% in 2004 and 60.8% in 2010 (http://www.cdc.gov/nchs/nhis/quest_data_related_1997_forward.htm). The reasons for this increasing rate of non-participation could be summarized as an overwhelming number of requests for study participation, a general decrease in volunteerism, distrust in science, and increasing complexity of studies that require lengthy consent forms or involve complicated procedures ([3] for review).

One major concern associated with non-participation is *non-response bias*, which refers to systematic errors in data that occur in a study when the reasons for (non)-participation are somehow linked to the main outcomes of interest [1; 3; 5; 6]. Researchers have tried numerous different ways to enhance response rates in attempts to minimize non-response bias and increase the representativeness of study samples. One such means is to utilize a variety of survey data collection methods within a single study [2; 3]. With a mixed methods approach, otherwise eligible individuals who fail to respond to one survey method (e.g., telephone interviews) are subsequently given the opportunity to respond via another (e.g., written questionnaires). However, problems with mixed methods designs may arise when responses differ as a function of the survey method selected [2; 3].

Consequently, the main aims of the present study were 1) to assess whether response rates vary by survey method (telephone vs. mail); 2) to evaluate whether the main outcome (in our case, the prevalence of children with special health care needs; CSHCN) is influenced by survey method; and 3) to identify reasons for non-participation.

METHODS

The 'National Survey of Children with Special Health Care Needs in Switzerland' (approved by the ethics committee of the University of Zurich) used a national representative sample of 16,496

children, ages 9-14 years, drawn from a canton/municipality-based population [7]. The main goal of the survey was to estimate the nationally prevalence of children with special health care needs using the 'CSHCN Screener' with parents as proxy respondents [8]. The survey primarily was conducted by computer-assisted telephone interviews. However, for those parents whose telephone numbers could not be found (2,859) or who could not be reached by telephone (1,340), a written screening questionnaire and information letter were sent by mail. On the written questionnaire, the parents were asked to provide their telephone number. If they did, they were contacted again by telephone. If the parents did not return the questionnaire within approximately one month, a reminder and second questionnaire were sent. Telephone interviews and questionnaires were available in all three official Swiss languages (German, French and Italian). The reason(s) parents refused to participate could only be assessed among those parents who were contacted by telephone. Multiple answers were permitted.

RESULTS

Altogether, 10,830 parents (or other caretakers) responded to the survey (9,371 by telephone interview; 1,044 by written questionnaire; 415 by written questionnaire *and* telephone interview). The overall response rate was 65.7%. For those parents whose telephone number was found and who were reached (12,297), the response rate was 76.2% (9,371/12,297). For those parents to whom the written questionnaires were sent (parents whose telephone numbers were not found initially (2,859) and those parents whose telephone numbers were found but were unable to be reached (1,340)), the response rate was 34.4% (1,459/4,199). Hence, those parents who were contacted by mail were more likely *not to participate* in the survey (OR=6.0[5.6-6.5]). Furthermore, parents of non-Swiss children were more likely *not to participate* in the survey relative to parents of Swiss children (non-participation rates: 46.2% vs. 22.8%; OR=2.9[2.6-3.2]). Due to the high association between the mode of data collection (telephone vs. written) and nationality (contacted by telephone: Swiss parents = 79.2% vs. non-Swiss parents = 57.6%; χ^2_1=409.043; p<.001), we included both factors in a logistic regression model to adjust for potential confounding. In this model, both factors remained significantly associated with

non-participation. However, the adjusted odds ratio for non-participation declined to 3.6[3.2-4.1] for those parents to whom the questionnaires were sent and to 2.4[2.1-2.7] for the parents of non-Swiss children.

Besides looking at response rates, we also assessed whether actual responses varied as a function of survey mode and found that the prevalence of CSHCN did not differ between those who responded by telephone and those who responded by mail (13.9% vs. 13.2%; χ^2_1=0.388; p=.534).

We further examined the reasons for non-participation among 1,800 parents who actively refused to participate, among whom 79.6% provided their reason(s) for non-participation (Table A.1). The most frequently mentioned reasons for non-participation were ‘did not want to provide information about their children’ (23.5%) and ‘lack of interest in the study topic’ (16.2%). We further examined the reasons for non-participation by nationality. Significant differences in reasons for non-participation were identified between parents of Swiss and non-Swiss children. For example, versus parents of non-Swiss children, significantly more parents of Swiss children mentioned 1) 'did not believe that data protection was ensured' (8.1% vs. 1.6%) and 2) 'lacked trust in the study/research' (6.1% vs. 1.6%) as their reasons for non-participation. On the other hand, significantly more parents of non-Swiss children mentioned: 'lack of interest in the study topic' (23.6% vs. 15.6%) and 'were concerned that the interview would be overheard by the telephone institute supervisor' (1.6% vs. 0.3%) as their reasons for nonparticipation.

Table A.1: Reasons for non-participation in the National Survey of Children with Special Health Care Needs in Switzerland (conducted in 2010/2011)

	Total sample *N* = 1,800	Parents of Swiss children *N* = 768	Parents of Non-Swiss children *N* = 123	χ^2_1
Did not want to provide information about their children	23.5%	22.0%	25.2%	.623
Lack of interest in the study topic	16.2%	15.6%	23.6%	4.814*
No time	8.0%	9.8%	12.2%	.689
Felt overwhelmed by telephone inquires	7.3%	6.5%	7.3%	.112
Felt that the topic was too personal	6.7%	6.5%	8.9%	.984
Did not believe that data protection was ensured	6.6%	8.1%	1.6%	6.609**
Lacked trust in the study/research	5.2%	6.1%	1.6%	4.120*
Personal issues	1.5%	1.3%	0.8%	.208
Did not want to provide information over the telephone	1.3%	0.8%	2.4%	2.914
Did not want to participate in any kind of survey as a matter of principal	0.7%	0.8%	0%	.967
Were concerned that the interview would be overheard by the telephone institute supervisor	0.4%	0.3%	1.6%	4.424*
Considered the interview too long	0.1%	0%	0%	-
Other reason(s)	11.8%	10.8%	9.8%	.123

Note: Reasons for non-participation given by refusing parents reached by telephone (multiple answers permitted).

Of the total 1,800 refusing parents, 1,433 (79.6%) provided reasons for their non-participation. Information on nationality was not available for all children; therefore, the *Ns* of parents of Swiss or non-Swiss children do not add up to 1,800. * = $p<.05$; ** = $p<.01$

DISCUSSION

Our response rate (65.7%) was similar to that of other Swiss national surveys (e.g., Swiss Health Survey; [9] and to that of U.S. surveys that have used the same instrument [10]. The response rate was much lower among those parents who were contacted by mail than those contacted by telephone. However, the results should be interpreted with caution, because parents were not randomly assigned to the mail versus telephone survey. It is possible that those parents whose telephone numbers could not be found in the public directory were more protective of their privacy and, therefore, less likely to participate in the survey. However, other studies also have identified higher response rates with telephone interviews than mailed surveys (e.g., [11]). Furthermore, more telephone numbers were found among parents of Swiss children than among parents of non-Swiss children. It is possible that more non-Swiss families choose not to list their telephone number in the public directories or that they just have mobile telephone numbers.

The finding that the response rate was higher among parents of Swiss than non-Swiss children could be attributable to language/comprehension problems. Furthermore, one of the frequently mentioned reasons for non-participation among parents of non-Swiss children was their lack of interest in the study topic that could be partially attributed to language/comprehension problems, because they might not completely understand the invitation letter that we sent them to introduce the study.

The finding that the prevalence of CSHCN did not differ by data collection mode is indicative of the usefulness of mixed survey methods: such an approach could be justified to increase participation in a study to assess CSHCN without introducing bias secondary to data collection method.

Lastly, it was demonstrated that a common theme percolating through several of the main refusal reasons was *distrust in science* ('did not want to provide information about their children', 'did not believe that data protection was ensured', 'lacked trust in the study/research', 'did not want to provide information over the telephone', 'were concerned that the interview would be overheard by the telephone institute supervisor'). Despite our efforts to address the trust issue, such as giving detailed information about the study and assuring all potential-participants about data protection, the problems remained. Other common reasons for non-participation included: A) a *lack of interest in the study*

topic (e.g. parents did not feel it was important to participate in the study when their child was healthy); B) *time constraints* ('no time', 'considered the interview too long'); C) *a rising number of study requests prompting a general refusal of all surveys* ('felt overwhelmed by telephone inquires', 'did not want to participate in any kind of survey as a matter of principal'); and D) *personal issues* ('felt that the topic was too personal', 'personal issues').

CONCLUSIONS

The present study demonstrates that a mixed survey methodology approach – combining telephone interviews and mailed written questionnaires – can be used to increase participation in surveys without influencing study outcomes. To enhance survey participation rates in the future, it will be especially important to increase the general population's overall trust in science (e.g., by advocating public information campaigns to increase science knowledge, and providing more detailed study introductions including how data will be handled to ensure data protection). In particular, greater efforts must be made to encourage and motivate parents of foreign nationalities to participate in studies (e.g., by providing additional language options to reduce non-participation secondary to comprehension difficulties).

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Table A.2: Group comparisons for the self- and parent-reported KIDSCREEN-27 scores between CSHCN with mental health problems with and without medication

	CSHCN mental: no medication mean (SD)	CSHCN mental: medication mean (SD)	t-test			ES (95% CI)
			t	df	*p*	
Parent						
Physical well-being	71.93 (16.72)	72.64 (17.06)	.459	508	.647	-0.04 (-0.22, 0.14)
Psychological well-being	74.50 (13.57)	72.90 (15.00)	-1.249	517	.212	0.11 (-0.06, 0.29)
Autonomy & parent relation	73.45 (13.54)	73.09 (13.92)	-.290	506	.772	0.03 (-0.15, 0.21)
Social support & peers	63.97 (21.08)	64.07 (21.56)	.050	507	.960	0 (-0.18, 0.17)
School environment	65.94 (17.39)	64.77 (19.58)	-.705	518	.481	0.06 (-0.11, 0.24)
Total HRQOL score	70.63 (11.94)	70.08 (13.07)	-.488	502	.626	0.04 (-0.14, 0.22)
Child						
Physical well-being	72.70 (17.08)	74.14 (16.88)	.864	443	.388	-0.08 (-0.28, 0.11)
Psychological well-being	80.99 (15.10)	79.57 (14.25)	-.994	452	.321	0.10 (-0.09, 0.29)
Autonomy & parent relation	77.86 (16.10)	77.15 (15.54)	-.450	434	.653	0.04 (-0.15, 0.24)
Social support & peers	74.51 (22.87)	76.75 (21.26)	1.030	448	.304	-0.10 (-0.29, 0.09)
School environment	72.38 (18.50)	72.89 (18.91)	.278	446	.781	-0.03 (-0.22, 0.16)
Total HRQOL score	76.57 (13.16)	76.14 (13.75)	-.328	437	.743	0.03 (-0.16, 0.23)

Note: *CI*: confidence interval; *CSHCN mental*: children with special health care needs with mental health problems; *ES*: effect sizes; *HRQOL*: health-related quality of life. The number of subjects (*N*) varies between the subscale and total HRQOL scores due to missing data. The largest *N* for CSHCN with mental health problem that were not on medication was 325 for the parent-ratings and 282 for the children-ratings. The largest *N* for CSHCN with mental health problem that were on medication was 198 for the parent-ratings and 172 for the children- ratings; effect sizes are designated as small (0.20), medium (0.50), and large (0.80)

Table A.3: Simple linear regression analyses (independent variables: presence of a mental or physical health constraint; dependent variable: parent- and child-reported health-related quality of life scores)

	Physical well-being		Psychological well-being		Autonomy & parent relation		Social support & peers		School environment		Total HRQOL	
	Parent	Child	Parent	Child	Parent	Child	Parent	Child	Parent	Child	Parent	Child
	β	β	β	β	β	β	β	β	β	β	β	β
Health status												
Controls (reference)												
CSHCN mental	-.196***	-.193***	-.318***	-.206***	-.170***	-.180***	-.208***	-.177***	-.373***	-.226***	-.632***	-.264***
CSHCN physical	-.269***	-.227***	-.176***	-.113***	-.075*	-.034	-.155***	-.115***	-.071*	-.036	-.182***	-.105***

Note: *CSHCN mental*: children with special health care needs with mental health problems; *CSHCN physical*: children with special health care needs with physical health problems; *HRQOL*: health-related quality of life

Both significant and non significant standardized betas are reported; * = $p \leq .05$; ** = $p \leq .01$; *** = $p \leq .001$

Table A.4: Group comparisons for self- and parent-reported KIDSCREEN-27 scores between CSHCN with mental health problems that were classified based on method 1 (main health problem) or method 2 (fifth item of the CSHCN Screener)

	Classified with method 1 mean (SD)	Classified with method 2 mean (SD)	t-test			ES (95% CI)
			t	df	*p*	
Parent						
Physical well-being	72.17 (16.87)	73.81 (14.60)	- .256	508	.798	-0.10 (-0.84, 0.65)
Psychological well-being	73.80 (14.14)	82.74 (11.39)	-1.543	517	.123	-0.63 (-1.44, 0.17)
Autonomy & parent relation	73.26 (13.68)	77.55 (13.48)	-.825	506	.410	-0.31 (-1.06, 0.43)
Social support & peers	63.81 (21.28)	78.57 (11.89)	-1.831	507	.068	-0.70 (-1.45, 0.05)
School environment	65.41 (18.22)	71.43 (20.68)	-.867	518	.387	-0.33 (-1.08, 0.42)
Total HRQOL score	70.33 (12.33)	77.50 (13.39)	-1.526	502	.128	-0.58 (-1.33, 0.16)
Child						
Physical well-being	73.21 (17.09)	76.66 (6.09)	-.452	443	.652	-0.20 (-1.08, 0.68)
Psychological well-being	80.36 (14.84)	88.57 (4.66)	-1.236	452	.217	-0.56 (-1.44, 0.33)
Autonomy & parent relation	77.54 (15.94)	81.43 (8.15)	-.544	434	.587	-0.25 (-1.13, 0.64)
Social support & peers	75.34 (22.36)	76.25 (15.56)	-.091	448	.928	-0.04 (-0.92, 0.84)
School environment	75.49 (18.68)	80.00 (14.25)	-.896	446	.371	-0.24 (-1.12, 0.64)
Total HRQOL score	76.36 (13.42)	80.50 (7.58)	-.688	437	.492	-0.31 (-1.19, 0.57)

Note: *CI*: confidence interval; *CSHCN*: children with special health care needs; *ES*: effect sizes; *HRQOL*: health-related quality of life

The number of subjects (*N*) varies between the subscale and total HRQOL scores due to missing data. The largest *N* for CSHCN with mental health problem classified by method 1 was 513 for the parent-ratings and 449 for the children-ratings. The largest *N* for CSHCN with mental health problem classified by method 2 was 7 for the parent-ratings and 5 for the children-ratings; effect sizes are designated as small (0.20), medium (0.50), and large (0.80)

Table A.5: Group comparisons for self- and parent-reported 'Strength and Difficulties Questionnaire' scores

				Kruskal-Wallis test			Effect sizes (95% CI)		
	CSHCN mental mean (SD)	CSHCN physical mean (SD)	Control mean (SD)	*H*	df	*p*	CSHCN mental vs. CSHCN physical	CSHCN mental vs. controls	CSHCN physical vs. controls
Parent									
Emotional	3.38 (2.43)[a]	2.18 (2.09)[b]	1.54 (1.68)[c]	205.829	2	$p<.0005$	0.52 (0.38, 0.66)	0.91 (0.79, 1.03)	0.35 (0.22, 0.46)
Conduct	2.53 (2.00)[a]	1.54 (1.44)[b]	1.34 (1.34)[b]	138.037	2	$p<.0005$	0.55 (0.41, 0.69)	0.72 (0.61, 0.84)	0.15 (0.02, 0.07)
Hyperactivity	4.75 (2.56)[a]	2.55 (2.18)[b]	2.33 (1.92)[b]	296.937	2	$p<.0005$	0.91 (0.76, 1.05)	1.10 (0.98, 1.22)	0.11 (-0.02, 0.18)
Peer	2.69 (2.46)[a]	1.88 (2.09)[b]	1.27 (1.56)[c]	118.963	2	$p<.0005$	0.35 (0.21, 0.49)	0.72 (0.60, 0.83)	0.35 (0.22, 0.31)
Prosocial	7.60 (1.97)[a]	8.23 (1.83)[b]	8.20 (1.70)[b]	37.798	2	$p<.0005$	-0.33 (-0.47, -0.19)	-0.33 (-0.44, -0.22)	0.02 (-0.11, -0.21)
Total	13.36 (6.56)[a]	8.14 (5.32)[b]	6.48 (4.59)[c]	373.006	2	$p<.0005$	0.85 (0.71, 1.00)	1.25 (1.13, 1.37)	0.34 (0.21, 0.16)
Child									
Emotional	3.03 (2.28)[a]	2.39 (2.01)[b]	2.05 (1.86)[b]	54.325	2	$p<.0005$	0.29 (0.14, 0.44)	0.48 (0.36, 0.60)	0.18 (-0.26, 0.51)
Conduct	2.44 (1.71)[a]	1.68 (1.30)[b]	1.61 (1.29)[b]	76.324	2	$p<.0005$	0.49 (0.33, 0.64)	0.56 (0.44, 0.68)	0.05 (-0.65, 0.12)
Hyperactivity	4.56 (2.23)[a]	3.26 (1.92)[b]	3.26 (2.08)[b]	101.234	2	$p<.0005$	0.61 (0.46, 0.77)	0.61 (0.49, 0.73)	0.00 (-0.77, 0.02)
Peer	2.61 (2.25)[a]	2.01 (1.97)[b]	1.56 (1.58)[c]	65.733	2	$p<.0005$	0.28 (0.13, 0.43)	0.56 (0.44, 0.68)	0.26 (-0.59, 0.19)
Prosocial	7.86 (1.81)[a]	8.35 (1.57)[b]	8.38 (1.52)[b]	23.005	2	$p<.0005$	-0.28 (-0.43, -0.13)	-0.32 (-0.44, -0.20)	-0.02 (-1.00, -0.21)
Total	12.64 (5.76)[a]	9.34 (4.62)[b]	8.49 (4.71)[c]	153.433	2	$p<.0005$	0.62 (0.46, 0.77)	0.81 (0.68, 0.93)	0.18 (0.81, 0.00)

Note: *CI*: confidence interval; *CSHCN mental*: children with special health care needs with mental health problems; *CSHCN physical*: children with special health care needs with physical health problems; *SDQ*: Strenght and Difficulties Questionnaire. SDQ scales: *emotional*: emotional symptoms; *conduct*: conduct problems; *peer*: peer problems; *prosocial*: prosocial behavior; *total*: total difficulties score. Higher SDQ-scores indicate more problems (exception: prosocial behavior). The number of subjects (*N*) varies between the subscale and total HRQOL scores due to missing data. The largest *N* for parent-ratings was 531 for CSHCN with mental health problems, 323 for CSHCN with physical health problems, and 743 for controls. The largest *N* for child-ratings was 458 for CSHCN with mental health problems, 278 for CSHCN with physical health problems and 682 for controls; subgroups with different superscripts are significantly different ($p<0.05$ (Bonferroni adjusted) with Mann-Whitney *post hoc* test); non-parametric tests were used because of non-normally distributed variables and inhomogeneous variances; effect sizes are designated as small (0.20), medium (0.50), and large (0.80)

Exploratory qualitative analyses

At this point, a short overview about the comments that were made about the child questionnaires is provided in Table A.6 (the sample used here is similar to the one described in Chapter 3). Analogous analyses could be conducted on the comments of parents.

Table A.6: Classification of the comments of children provided on the health-related quality of life questionnaires

Topic	Example
Super-ordinate positive statements	Life is great!
Statements on the questionnaire/survey	
Filling-out the questionnaire	"Mum has read the questions and explained them to me."
Positive statements	"It was fun to fill out the questionnaire. If you ever have such a thing again: please send it to me."
Negative statements	"The questions are not clear and precise."
Holidays	"I was at a Scouts camp over the last two weeks."
Questions related to the survey	"Why did I have to answer these questions?"
Peers	
Positive statements	"I have a lot of friends."
Negative statements	"My schoolfellows never liked me."
Family/experiences regarding family	
Positive statements	"I like my parents!"
Negative statements	"I am sad because I see my dad only rarely."
School	
Positive statements	"I feel very good in school."
Negative statements	"School is very stressful for me."
Health condition	
Mentioning the health condition	"I am hyperactive."
Statements related to medication	"When I take medication, I do not have any problems."
Positive reference to the health condition	"I am happy despite being dyslexic."
Negative reference to the health condition	"The dyslexia is a part of my life, and this is difficult for me."
Others	
Statements about pocket money	"I do not receive pocket money."
Others	"I do not know."

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CURRICULUM VITAE

DEY Michelle

Working Contact
University of Zurich
Institute of Social- and Preventive Medicine
Hirschengraben 84
8001 Zurich
Phone: ++41 44 634 54 22
Fax: ++41 44 634 49 86
Email: michelle.dey@uzh.ch

EDUCATION

2009 – 2012 Doctoral student at the Institute of Social and Preventive Medicine, University of Zurich, Switzerland
Prof. M. A. Landolt, Ph.D.; Prof. G. Bodenmann, Ph.D.; M. Mohler-Kuo, Sc.D.
Conducting the NS-CSHCN-CH, wrting the Ph.D. thesis in Psychology (title: HRQOL among children with mental health problems)

2007 Master of Science in Psychology, Psychopathology (infancy and adolescence), Social and Preventive Medicine at the University of Zurich, Switzerland
Master thesis (translated): Learning and memory functioning of children suffering from attention-deficit/hyperactivity disorder (ADHD)

2001 Teacher's certificate, Lehrerseminar Köniz-Lerbermatt, Berne, Switzerland

WORK EXPERIENCE

2007 – 2009 Project manager/assistant at Pro Juventute, Zurich, Switzerland

Evaluation of the effectiveness of Pro Juventute's 'letters to parents' (containing information on such topics as the development, upbringing and diet of toodlers) on a national level, planning and implementation of further products for parents, fundraising

2005 – 2006 Research assistant at the Department of Child and Adolescent Psychiatry, University of Zurich, Switzerland

R. Drechsler, Ph.D.

Conducting neurofeedback-sessions with children suffering from attention-deficit/hyperactivity disorder (ADHD), support of studies on the efficacy of neurofeedback as well as group therapy among children with ADHD

2004 – 2005 Research assistant at the Department of Psychology, Neuropsychology, University of Zurich, Switzerland

Prof. L. Jäncke, Ph.D.

Tutoring undergraduate students, neuropsychological evaluation of patients and preparation of neuropsychological reports, conducting and publishing a driving simulator study about the influence of music on the driving behavior

LANGUAGES

German: Native

English: Advanced

French: Basic

PUBLICATIONS

Dey, M., Landolt, M. A., Mohler-Kuo, M. (2012). Assessing parent-child agreement in health-related quality of life among three health status groups. *Social Psychiatry and Psychiatric Epidemiology*, doi: 10.1007/s00127-012-0556-z.

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Dey, M. & Mohler-Kuo, M. (2011). *Children with special health care needs in Switzerland. Preliminary findings of a national survey*. Presentation at the Swiss Public Health Conference 2011, Basel, Switzerland, 25th – 26th August.

Dey, M. & Mohler-Kuo, M. *Reasons for parents not participating in a national screening survey to identify children with and without special health care needs*. Poster presentation at the Swiss Public Health Conference 2011, Basel, Switzerland, 25th – 26th August.

Mohler-Kuo, M., **Dey, M.** & Zellweger, U. (2011). *Barriers and satisfaction of health care utilization among children with special health care needs*. Poster presentation at the Swiss Public Health Conference 2010, Nottwil, 9th – 10th September.

Mohler-Kuo, M., **Dey, M.** & Zellweger, U. (2010). *Health-related quality of life among children with special health care needs*. Poster presentation at the 20th IUHPE World Conference on Health Promotion, Geneva, Switzerland, 11th – 15th July 2010.